CASE REPORT

Abdominal Cocoon: A Rare Case of Intestinal Obstruction

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Abstract

Abdominal cocoon [encapsulating peritoneal sclerosis (EPS)] is a rare cause of intestinal obstruction characterized by the formation of a fibrocollagenous peritoneal membrane that encases abdominal viscera, mainly small intestinal loops. It is believed to be a result of a chronic intra-abdominal fibro-inflammatory process that results in the formation of marbled, thickened leathery cocoon-like fibroconnective tissue sheets that cover, fix, and ultimately constrict the gut compromising its motility. EPS can be primary (idiopathic) or secondary to several other causes like long-term peritoneal dialysis, postrenal transplantation, abdominal tuberculosis (TB), peritoneal shunts, etc. The etiology of the primary EPS is unknown, and diagnosis is mostly made after exploratory laparotomy and histopathological analysis of the sac. Surgical exploration with resection of thick membranes and associated adhesions is the mainstay of the treatment. The condition has a good postoperative outcome, provided it is diagnosed and managed early. We herein report an unusual case of a 43-year-old patient who developed intestinal obstruction-like symptoms with vomiting and abdominal pain. Computed tomography (CT) scan revealed typical findings of the abdominal cocoon which was later confirmed at surgery. To our best knowledge, only a few such cases have been reported so far.

Keywords: Abdominal cocoon, Encapsulating peritoneal sclerosis, Intestinal obstruction.

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Introduction

Encapsulating peritoneal sclerosis is an uncommon etiology for intestinal obstruction, characterized by the formation of a fibrocollagenous membrane that encapsulates intestinal loops. It is believed to be an end result of a chronic intra-abdominal fibro-inflammatory process that leads to the formation of marbled, leathery cocoon-like fibroconnective tissue sheets that encapsulate and eventually constrict the gut, affecting its motility. It was first narrated by Foo et al.[1] Incidence is equal in both genders and occurs over a vast range of age-groups.[2] Clinically it presents with features of altered intestine motility initially, to overt symptoms of bowel obstruction as soon as complete sclerosis sets in.

Encapsulating peritoneal sclerosis may be primary or secondary to cases of long-term peritoneal dialysis, post-kidney transplant, peritoneal TB, etc.[3] Diagnosis is, in general, made after exploratory laparotomy and histopathological study of the sac. Surgical exploration, resection of membranes, and associated adhesive sac are the mainstay of management. EPS has a good postoperative outcome, provided it is identified and treated early.

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Case Description

A 43-year-old man was admitted to our hospital with acute onset continuous diffuse abdominal pain since 8 hours, associated with 3–4 episodes of vomiting. The patient told that he had similar complaints in the last 1 year which required hospital admissions and were relieved by conservative management. The patient is a known case of hypothyroidism and is on thyroxine hormone supplement (75 µg/d) since 5 years. He did not have a history of any other chronic conditions like TB, or any history of abdominal surgery. On admission, the patient had bradycardia. On physical examination, the abdomen was soft, bowel sounds were sluggish with diffuse tenderness, and some vague mass was palpable in right lumbar and periumbilical region, the nature of which was not clinically well appreciable. A digital rectal examination did not reveal any abnormality and other systemic examinations were unremarkable. Basic laboratory parameters were within normal limits except for raised thyroid-stimulating hormone levels of 24 and slightly lower free T4 levels.

Contrast-enhanced computed tomography (CECT) abdomen showed features suggestive of encapsulation of small bowel loops by peritoneal sac from the duodenojejunal junction to distal ileum, with mildly dilated small intestinal loops suggestive of small intestinal obstruction. A provisional diagnosis of EPS (abdominal cocoon) was made based on clinical history, physical examination, and radiological findings.

During exploratory laparotomy, a small bowel from DJ flexure up to the ileocecal junction (ICJ) was encased in thick white-colored fibrous tissue with extensive adhesions. Stomach and large bowels were spared. Extensive adhesiolysis was done and the small bowel from DJ flexure to ICJ encased in the fibrous membrane was released

and fibrous tissues excised. Small bowel from mid-jejunum to ICJ had gangrenous changes and right hemicolectomy was done and specimens were sent for histopathological examination. Specimens showed walls of intestines with mucosal and submucosal edema with hemorrhagic necrotic changes. Serosa showed adherent exudates and a thick band of fibrous tissue. Visceral peritoneum showed proliferation of fibroconnective tissue and inflammatory infiltrates composed of scattered lymphocytes, plasma cells, and few neutrophils. There were no calcifications/foreign body granulomas/giant cells. No malignant cells were seen.

Postoperatively, he was given total parenteral nutrition and he tolerated it well. The patient was followed up postoperatively for 15 days and his postoperative recovery was satisfactory without any complications (Figs 1 to 3).

Discussion

Abdominal cocoon syndrome/sclerosing encapsulating peritonitis is a rare disease of the peritoneum and almost invariably presents as an acute or subacute intestinal obstruction with or without a mass, which is usually diagnosed incidentally at laparotomy. It is often confused with congenital peritoneal encapsulation which is a rare developmental anomaly in which part or the entire small intestine is encased in an accessory sac derived from the yolk sac.[4] Small bowel is thus covered by the dorsal mesentery which usually forms the transverse mesocolon, hence forming the characteristic accessory peritoneal sac. This membrane is free from underlying intestinal loops and the histology of the sac is mainly mesothelial with or without chronic inflammation or fibrosis.[5,6]

Idiopathic/primary EPS must be differentiated from that secondary to other conditions. Idiopathic EPS also called abdominal cocoon is a rare condition of unknown etiology. It is characterized by a thick grayish-white fibrotic membrane, partially or totally covering the small intestine. Idiopathic EPS manifests with features of recurrent acute or chronic small intestinal obstruction due to entangling and/or constriction of bowels within the compressing cocoon. It can manifest as a lump in the abdomen also due to dilated small intestinal loops.[1,7] Radiologically, CECT abdomen is useful in the diagnosis of idiopathic EPS and abdominal X-rays are usually nonspecific. CECT images show small bowel loops being encapsulated by fibrous tissues. These findings are not pathognomonic but help in the diagnosis and management of this condition. Idiopathic EPS is usually a diagnosis of exclusion.[8]

A definitive diagnosis of idiopathic EPS is done at exploratory laparotomy where the intestinal loops are found in twisted form with or without adhesions, covered with a thick glistening membrane encasing the small intestinal loops. In our case, the small bowel from DJ flexure up to ICJ was encased in a thick white-colored fibrous membrane with extensive adhesions with sparing of the stomach and large bowel. The histopathological study of the sac usually shows different ranges of fibrosis and inflammatory changes.[7]

Treatment options of idiopathic EPS (abdominal cocoon) are mainly surgical and the most widely used surgical procedure is complete resection of the membrane with adhesiolysis. Resection and anastomosis are performed in a nonviable gut.[9] Other surgical approaches are releasing of adhesions, partial intestinal resection and anastomosis, total or subtotal excision of the sac, enterolysis, small intestinal intubation, etc., whenever necessary. Long-term postoperative prognosis is very good with minimal risk of recurrence. The most common surgical complications included intra-abdominal infections, enterocutaneous fistula, and bowel perforation.[10]

Successful conservative management has been reported with corticosteroids, immunosuppressants, and tamoxifen in sclerosing encapsulating peritonitis (SEP) secondary to continuous ambulatory peritoneal dialysis.[11]

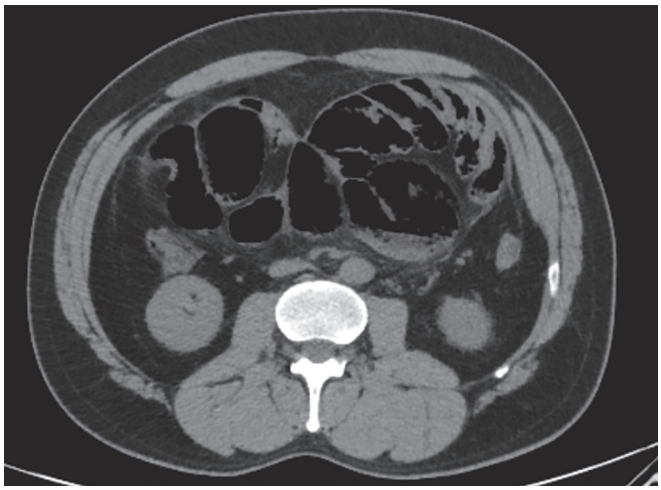

Fig. 1: CT axial image of abdominal cocoon

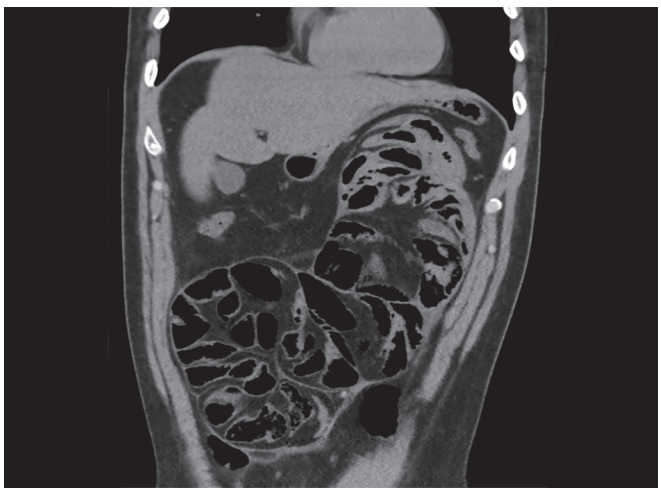

Fig. 2: Sagittal image of abdominal cocoon

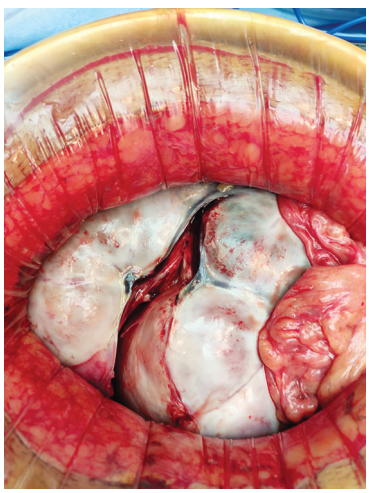

Fig. 3: Surgical image of cocoon

Conclusion

The aim of this study is to raise awareness about this condition. Most cases of abdominal cocoon syndrome (ACS) are diagnosed incidentally during laparotomy. A high index of clinical suspicion (recurrent episodes of non-strangulating small intestinal obstruction in absence of the common etiologies) combined with imaging modality like CT scan may help to make a preoperative diagnosis. We seek to acquaint clinicians with this uncommon entity and highlight the importance of its early diagnosis to prevent unnecessary bowel resections and worst outcomes.

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