

Abdominal Cocoon in an Adolescent Male

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ABSTRACT

Abdominal cocoon is an uncommon peritoneal condition characterized by partial or complete encasement of small bowel by a thick rind of fibrous tissue and adhesions. Only few cases of this disease have been reported in world literature. The etiology of this condition is unknown, and most often it is found in adolescent girls from tropical or subtropical countries, and usually diagnosed incidentally on exploration. Surgery (membrane dissection and extensive adhesiolysis) is the treatment of choice. Here, we report a case of abdominal cocoon in a 20-year-old male patient, with a brief review of the literature.

KEYWORDS

Abdominal cocoon; adhesiolysis; intestinal obstruction

Citation

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INTRODUCTION

“Abdominal cocoon” is a rare condition of unknown etiology occurring mostly in adolescent girls living in tropical and subtropical regions.¹ It was first described by Foo et al.² It is characterized by the encasement of the small bowel by a fibro-collagenic membrane leading to its clustering, causing intestinal obstruction.¹ Only about 50 reports of this disease have been found in world literature.¹ Preoperative diagnosis of this entity is difficult, requires a high index of clinical suspicion and is usually incidentally diagnosed on laparotomy. Here we report a case of abdominal cocoon in a 20-year-old male patient.

CASE REPORT

A 20-years-old male presented in the emergency of our hospital with complaints of abdominal pain, vomiting, non-passage of stool and flatus and abdominal distension for last two days. He had intermittent episodes of abdominal pain and vomiting without abdominal distension for last one year, was empirically diagnosed to have abdominal

tuberculosis outside and kept on anti-tubercular treatment for last four months. There was no history of peritonitis, abdominal surgery or pulmonary tuberculosis. General physical examination revealed mild dehydration. His pulse was 110/minute, temperature 37.8°C, blood pressure 130/70 mmHg. There was no cyanosis or jaundice. No abnormalities of the chest or cardiovascular system were found. On per-abdominal examination, it was found to have predominant fullness in the lower half of the abdomen alongwith mild tenderness and rigidity. There was no hepatomegaly or splenomegaly and rectum was empty. The bowel sounds was exaggerated. His hemoglobin was 12.9 g%, total leukocyte count 16,765 cells/ml with 75% neutrophils, other blood chemistry and urine analysis were normal. The abdominal radiograph showed multiple air-fluid levels predominantly in the lower abdomen (Fig 1). A clinical diagnosis of mechanical small bowel obstruction was made provisionally, and emergency laparotomy was performed through a midline incision. Intra-operatively, it was found that the omentum and the small bowel loops were difficult to dissect from the parietal peritoneum,



Figure 1. A plain abdominal radiograph showing multiple bowel loops predominantly in the lower abdomen.

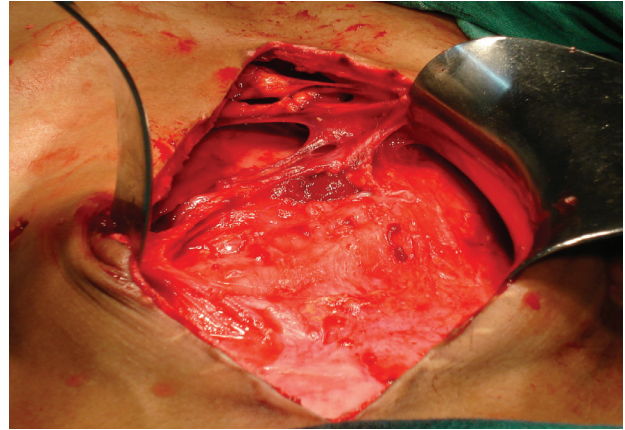


Figure 2. Laparotomy showing intimate adhesions of the bowel loops and omentum, making individual loops indistinguishable (abdominal cocoon).

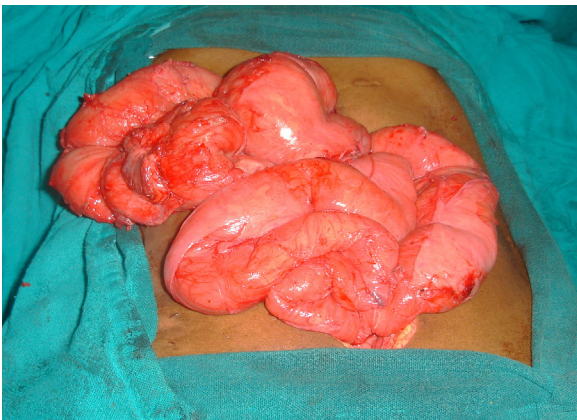


Figure 3. After adhesiolysis, small bowel loops are visible properly.

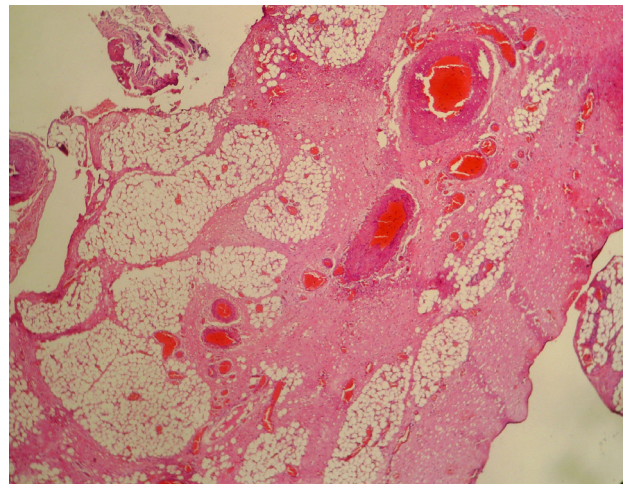


Figure 4. Photomicrograph of the omentum showing dilated and congested blood vessels and marked fibrosis (X10, H&E stain).

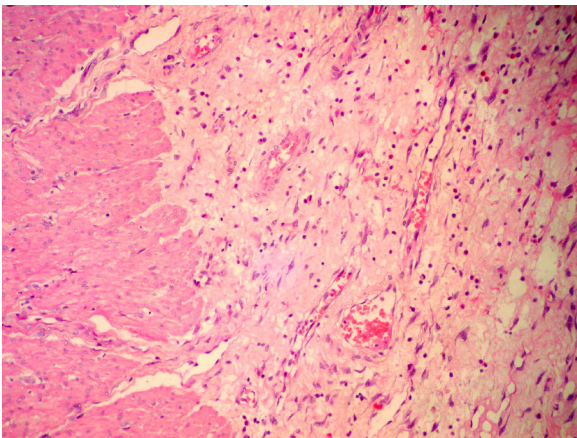


Figure 5. Photomicrograph of the omentum showing dilated and congested blood vessels and marked fibrosis (X10, H&E stain).

they were so intimately adherent to each other at the anterior aspect, that individual loops were almost indistinguishable, giving a suspicion of abdominal cocoon (Fig 2). A plane between the bowel loops was found with difficulty at one point, and using a combination of blunt and sharp dissection, adhesiolysis was done. There was

no perceptible separate membrane. (fig The whole small bowel was freed; during adhesiolysis, a large perforation occurred at proximal ileum, and after resection of about 20cm of the ileum, proximal ileostomy and distal mucous fistula was made. The resected bowel and the omentum were sent for histopathology. On microscopy, the omental tissue showed dilated and congested blood vessels and marked fibrosis; the serosa of the resected bowel showed thickening, mixed inflammatory cell infiltrates, consisting of neutrophils, eosinophils and lymphocytes alongwith fibrosis and congestion (Fig 4,5). A diagnosis of idiopathic sclerosing encapsulating peritonitis (abdominal cocoon) was established, due to intraoperative findings and by exclusion of other probable pathologies. The stoma started to function on 3rd postoperative day; he was orally given on 4th postoperative day, and was discharged on the 6th day. The ileostomy was closed six months after the initial operation, and he is on regular follow-up for the last one year without any bowel symptoms.

DISCUSSION

Abdominal cocoon is an uncommon peritoneal condition characterized by partial or complete encasement of small bowel by a thick rind of fibrous tissue and adhesions, causing clustering of the bowel and intestinal obstruction.¹ The disease primarily involves small bowel, but can extend to involve other organs like the large intestine, liver and stomach.^{1,3}

Abdominal cocoon can be idiopathic or secondary. Foo et al first described the idiopathic form, which is more common.² It generally affects adolescent girls, and is found to be more common in tropical and subtropical countries of the world, especially China, Malaysia, Singapore, Pakistan, India, Nigeria, Kenya, Saudi Arabia, Israel, and South Africa.⁴⁻⁶ Although its etiology is still unknown, subclinical primary viral peritonitis, as an immunological reaction to gynecological infections, or due to retrograde menstruation, and retrograde peritonitis via the fallopian tubes have been suggested for its pathogenesis, probably due to its increased associated incidence in women.² The secondary form is most commonly associated with chronic ambulatory peritoneal dialysis.^{7,8} It has also been described in association with prolonged practolol therapy, sarcoidosis, systemic lupus erythematosus, indwelling abdominal catheters (specifically ventriculoperitoneal and peritoneovenous shunts), orthotopic liver transplantation, liver cirrhosis, intraperitoneal instillation of drugs, recurrent peritonitis and tuberculous pelvic inflammatory disease.⁹⁻¹³

These patients usually present with features of acute or subacute small bowel obstruction, symptoms of chronic obstruction and weight loss, and/or pain associated with an abdominal lump.^{5,14} A pre-operative diagnosis is usually very difficult and requires a high index of clinical suspicion. Most cases are diagnosed intraoperatively during laparotomy performed for intestinal obstruction.

Plain radiographs of the abdomen may suggest features of intestinal obstruction. A preoperative diagnosis has been made in recent reports by a combination of barium follow-through and computed tomography of the abdomen.^{3,15,16} Barium meal follow-through may show delayed transit of contrast and clustering of the bowel loops in the pelvis.¹⁵ Pre-operative CT findings may include clumping of small bowel loops in the centre of the abdomen encased by a soft-tissue density mantle, peritoneal thickening and calcification, clumping of small bowel loops, and loculated fluid collections.^{3,16} Histopathological examination shows thickened vascular fibrocollagenous tissue, with or without chronic inflammatory reaction evidenced by lymphocytic and plasma cell infiltrates.

The treatment of abdominal cocoon has generally been described as dissection and adhesiolysis, which is usually sufficient. Resection is indicated only if the bowel is non-viable and grossly damaged during dissection, as in our case. Stoma has to be made in the most proximally located perforated region. Diagnostic laparoscopy and laparoscopic adhesiolysis has also been effective in the management of this disease.¹⁷ Though prognosis is said to be excellent, follow-up has also shown recurrence, some resolving on conservative management and others needing reoperation.^{6,14,18} Small bowel intubation has also been used with the hope of decreasing the recurrence as in postoperative adhesive obstruction.¹⁹

CONCLUSION

Abdominal cocoon is a rare condition, and the pre-operative diagnosis is usually difficult. Recurrent episodes of small intestinal obstruction not attributable to other causes combined with relevant imaging findings should point to suspicion of this disease. Surgery is important in its management. Careful dissection and adhesiolysis releasing the small intestine usually leads to recovery.

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