A Unusual Cause of Intestinal Obstruction: Abdominal Cocoon

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ABSTRACT

Abdominal cocoon syndrome is a rare cause of intestinal obstruction, which is difficult to diagnose preoperatively. We report a case of abdominal cocoon. A 47-year-old male patient was referred to the general surgery department with complaints of abdominal pain, distension, nausea, vomiting for one day. The abdominal CT examination detected dilated small intestinal loops clustered in the abdomen surrounded by a saclike membrane. During exploratory surgery, a capsular structure was identified in the lower quadrant with a regular surface that was solid-fibrous in nature. The combination of physical examination, imaging signs and the medical history, may be helpful in diagnosis.

Keywords: Abdominal cocoon, diagnosis, intestinal obstruction, surgery

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INTRODUCTION

Intestinal obstruction is seen frequently by surgeons all over the world. Commonly, the cause included adhesions, bands, inguinal hernia and tuberculosis. The 'cocoon' remains an obscure and relatively uncommon cause (1). 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, subactue or chronic bowel obstruction. It is predominantly reported among females from the tropical and subtropical regions (2). In this case report, we presented a case of intestinal obstruction in a male patient who was diagnosed with an abdominal cocoon and was managed successfully in our hospital.

CASE REPORT

A 47-year-old male patient was referred to the general surgery department with complaints of abdominal pain, distension, nausea, vomiting and constipation for one day. He had clinical history of several attacks of abdominal pain and nausea with spontaneous symptomatic relief over the lastyear. The patient had no history of previous abdominal operation and chronic systemic disease. On physical examination, there was obviously distension and general tenderness, especially prominent in lower regions of the abdomen, with a soft and mobile mass being palpated in the abdomen. Bowel sounds were a bit hyperactive, and rectal examination was normal. No hepatomegaly or splenomegaly was observed.

The abdominal radiography showed multiple air-fluid levels which were more prominent in the left lower quadrant (Figure 1, Abdominal radiography, multiple air-fluid levels are seen, which was more prominent in the left lower quadrant), and a provisional diagnosis of intestinal obstruction was considered.

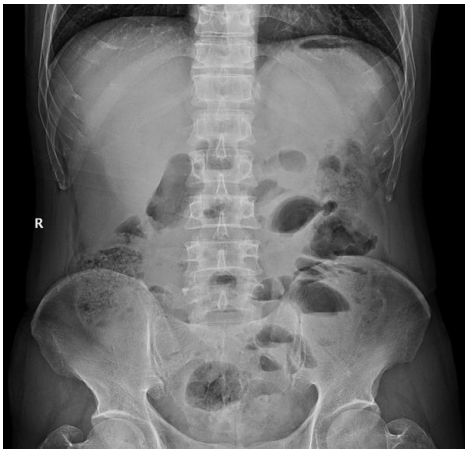

Fig.1: Abdominal radiography, multiple air-fluid levels are seen, which was more prominent in the left lower quadrant.

The CT scan of the abdomen showed multiple clumped and dilated small intestinal loops containing air-fluid levels and intestinal contents clustered in the right lower abdomen, which were surrounded by a thick, saclike membrane (Figure 2, CT scan showing a conglomerate of multiple intestinal loops surrounded by a thick sac-like membrane -white arrow, dilated small bowel loops and a small amount of ascitic fluid-black arrow, A and B:axial view; C: Coronal view; D:Sagittal view). There was a small amount of ascites in the pelvic region (Figure 2).

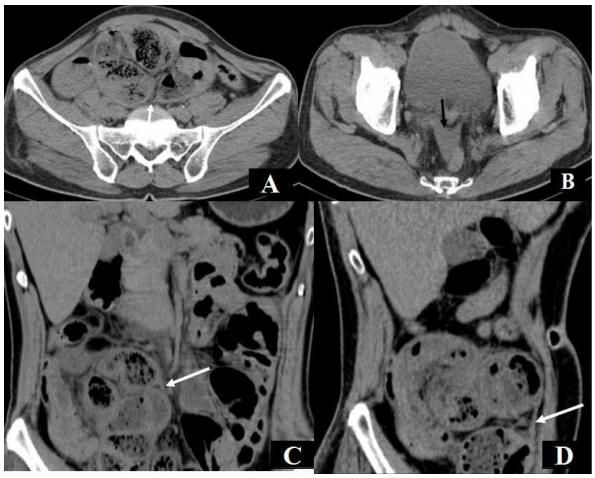

Fig.2: CT scan showing a conglomerate of multiple intestinal loops surrounded by a thick sac-like membrane (white arrow), dilated small bowel loops and a small amount of ascitic fluid (black arrow) (A and B:axial view; C: Coronal view; D:Sagittal view).

Ultrasound scan showed dilated and clumped bowel loops with reduced peristaltic activity, and an intestinal thick-walled mass, containing bowel loops. The laboratory examinations were normal, except for leukocytosis (16.000 mm^3).

The patient was hospitalized for emergency exploratory laparotomy. Intraoperative findings showed encapsulation of small bowel by a cocoon-like fibrous membrane (Figure 3), and intestinal wall edema was seen in the terminal ileum. Lysis of the membrane was carried out, loops were separated by dissection, and the adhesive loops of small bowel were freed. The freed small bowel segments were viable (Figure 3, Operative views: A, showing the ‘cocoon’ coverage of the small bowel. B, The cocoon was opened to release the contained small bowel loops, and the adherent loops of small bowel were freed. C, Complete excision of the sac and the release of entire small bowel). Other abdominal organs were normal.

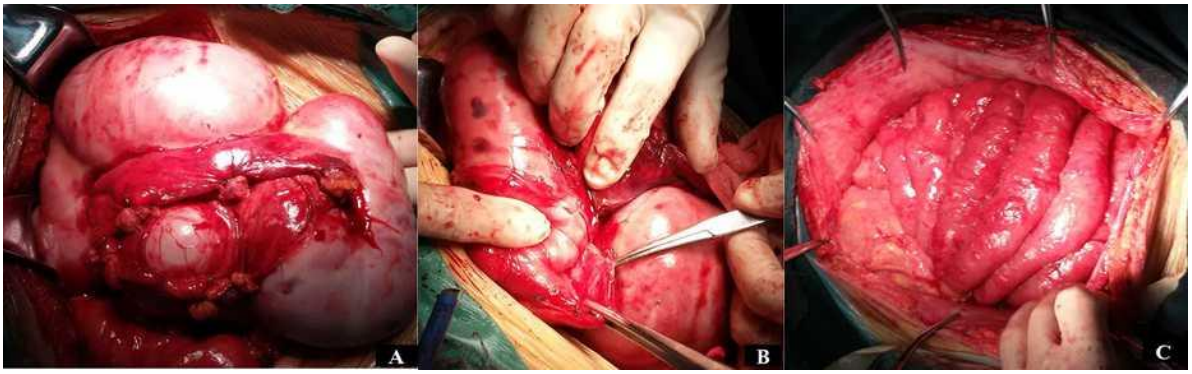

Fig.3: Operative views: (A) showing the ‘cocoon’ coverage of the small bowel. (B) The cocoon was opened to release the contained small bowel loops, and the adherent loops of small bowel were freed. (C) Complete excision of the sac and the release of entire small bowel.

The histological examination of the membrane revealed fibrous tissue focally lined by flatted mesothelial cells with scattered mononuclear inflammatory cell infiltrate and tissue culture fibroblasts. The patient recovered well and without developing recurrence at the follow-up four months later.

DISCUSSION

Since its first description in 1907 by Owtschinnikow, the abdominal cocoon has remained an uncommon cause of intestinal obstruction (acute, subacute or chronic intestinal obstruction) in which bowel loops of variable extent are encased within a fibrous sac (3). However, the term“abdominal cocoon” was first applied by Foo et al in 1978 (4). This is typically seen in young, adolescent females from the tropical and subtropical countries. Few cases of males have been diagnosed with abdominal cocoon. As yet, the underlying etiology is still uncertain. It could be primary or secondary. Primary condition is idiopathic, secondary condition is commonly seen

in the conditions that result in chronic asymptomatic peritonitis (such as: the use of practalol, peritoneal dialysis, endometriosis, and abdominal tuberculosis). Moreover, secondary causes accompanied by some diseases (systemic lupus erythematosus, familial Mediterranean fever, infections of the Fallopian tubes, retrograde menstruation, and gastrointestinal stromal tumours) have also been reported (2, 5-8). Yeniay et al. (2) reported that genetic factors may also play a role in the etiology. Even so, more work has to be done to elucidate the exact etiology of abdominal cocoon.

The preoperative diagnosis of abdominal cocoon is difficult since the clinical manifestations including abdominal pain, distension, nausea, abdominal fullness, vomiting, an abdominal mass and bowel obstruction, are nonspecific and variable between patients. Despite these difficulties, four main clinical features are suggested by Yip and Lee (8) for assisting preoperative diagnosis. First, a relatively young female patient without an obvious cause of bowel obstruction. Second, a past history of similar episodes with symptomatic relief. Third, presentation with symptoms suggestive of bowel obstruction but absence of cardinal symptoms such as distention. Fourth, presence of soft non-tender abdominal mass. Our case fits most of the above-mentioned features except for presence of the first and third features. All these clinical features allowed us to suspect the diagnosis of abdominal cocoon syndrome.

Imaging examinations play an important role in preoperative diagnosis of the disease, as shown in our case report. The abdominal radiography hinted a provisional diagnosis of intestinal obstruction. Multi-detector row computed tomography, especially contrast-enhanced computed tomography showed that clusters of intestinal loops were enclosed within a membrane-like sac,

bowel wall thickening, localized fluid collections. Ultrasound presented a thick-walled mass containing dilated and clumped bowel loops. These imaging features are suggestive of the diagnosis of abdominal cocoon. Explorative laparotomy and laparoscopy may improve the diagnostic accuracy when the membrane encased bowel loops are seen. Studies reported that the cocoon usually involves varying parts of the small intestine, but can extend on to cover the colon, stomach, liver and even spleen (9).

Surgery is the method of choice of symptomatic abdominal cocoon in almost all reported studies in the literature (8, 9). The principle is simply freeing the adhesions and excising the covering fibrous membrane on the small intestine carefully as much as possible. Intraoperative findings of our patient show coils of intestine covered with thick fibrous membrane and adhesions between the coils. Resection of encapsulating membrane will return bowel loops to peritoneal cavity. Breaking of adhesions needs to be done carefully, to prevent damage to serosal surface and perforation. In our case, encapsulating membrane was excised and adhesions were released. Resection of bowel was not required, because bowel loops were not strangulated.

In conclusion, we report a case of intestinal obstruction caused by abdominal cocoon which is a rarely seen condition, with preoperative diagnosis being difficult. The combination of physical examination and imaging features, and the knowledge of a careful medical history, may be helpful in diagnosis. The preoperative diagnosis requires a high index of suspicion, supported by clinical data and imaging findings indicative of the condition. In particular, CT imaging plays an important role in identifying these typical features related to the cause of abdominal cocoon,

although most cases are diagnosed at exploratory laparotomy. Suitable and early surgery and appropriate perioperative treatment can improve patient prognosis in this rare condition (10).

Conflict of interest

None declared.

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Ethical approval

Patient's consent was obtained and is available for review upon editor's request.

Authors’contribution

Yanguang Shen initiated the manuscript, prepared draft of the manuscript.Yusen Shi, Haining Cui, Rong Tu, Lanfang Su, Xiaohua Li, Xiaohua Zhou did literature review and participated in the discussion of the cases. Zhonghua Sun revised and approved the final draft.

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