CASE REPORT

An Unusual Cause of Intestinal Obstruction: Abdominal Cocoon

Y Shen[1], Y Shi[1], H Cui[2], R Tu[1], L Su[1], X Li[1], X Zhou[2], Z Sun[3]

ABSTRACT

Abdominal cocoon syndrome is a rare cause of intestinal obstruction, which is difficult to diagnose preoperatively. We here 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 and vomiting for 1 day. An abdominal computed tomography examination detected the dilated small intestinal loops clustered in the abdomen and surrounded by a sac-like membrane. During the 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 medical history may be helpful in the diagnosis.

Keywords: Abdominal cocoon, diagnosis, intestinal obstruction, surgery.

INTRODUCTION

Intestinal obstruction is seen frequently by surgeons all over the world. Commonly, the causes include adhesions, bands, inguinal hernia and tuberculosis. The 'cocoon' remains an obscure and a relatively uncommon cause (1). Abdominal cocoon is a rare condition that refers to the total or the partial encapsulation of the small bowel by a fibrocollagenous membrane or cocoon with local inflammatory infiltrate leading to acute, subacute or chronic bowel obstruction. It is predominantly reported among females from the tropical and subtropical regions (2). In this case report, we present a case of intestinal obstruction in a male patient who was diagnosed with an abdominal cocoon and was treated successfully in Hainan Medical College, Haikou, City of Hainan Province, China.

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 1 day. He had clinical history of many attacks of abdominal pain and nausea with spontaneous symptomatic relief during the previous year. The patient had no history of previous abdominal operation and chronic systemic disease. On his physical examination, there was obvious distension and general tenderness, especially prominent in lower regions of his abdomen, with a soft and mobile mass being palpated in his abdomen. His bowel sounds were a bit hyperactive, and his 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 (Fig. 1), and a provisional diagnosis of his intestinal obstruction was considered.

CT scan of the patient's abdomen showed multiple clumped and dilated small intestinal loops containing air-fluid levels and intestinal contents clustered in his right lower abdomen, which were surrounded by a thick, sac-like membrane (Fig. 2). There was a small amount of ascites in his pelvic region (Fig. 2).

Ultrasound scan showed dilated and clumped bowel loops with reduced peristaltic activity, and an intestinal thick-walled mass, containing bowel loops. His laboratory examinations were normal, except for leukocytosis (16 000 mm^3).

From: [1]Department of Radiology, Hospital Affiliated to Hainan Medical College, Haikou City of Hainan Province, China, [2]Department of General Surgery, Hospital Affiliated to Hainan Medical College, Haikou City of Hainan Province, China and [3]Discipline of Medical Imaging, Department of Imaging and Applied Physics, Curtin University, Perth, Australia.

Correspondence: Prof Y Shen, Department of Radiology, Hospital Affiliated to Hainan Medical College, Haikou, City of Hainan Province, 570102, China. Email: shenyanguang@163.com

 DOI: 10.7727/wimj.2015.244

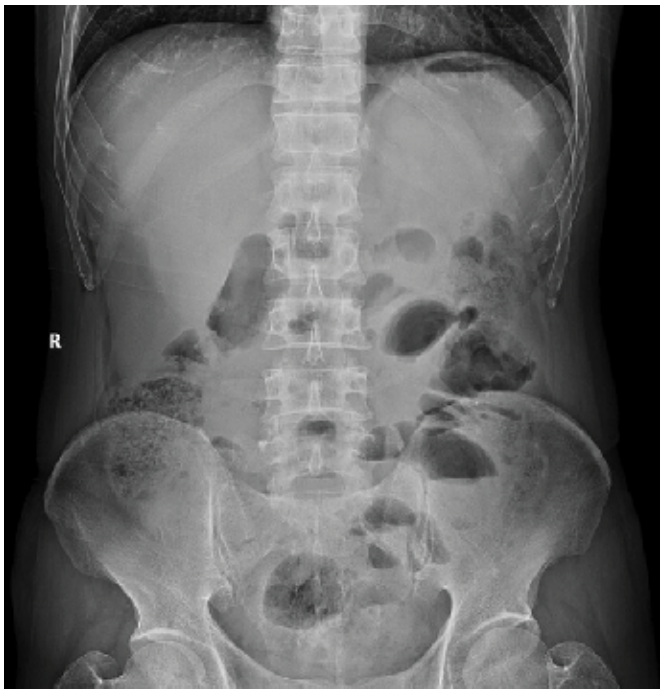

Fig. 1: Abdominal radiography shows multiple air-fluid levels, which were more prominent in the left lower quadrant.

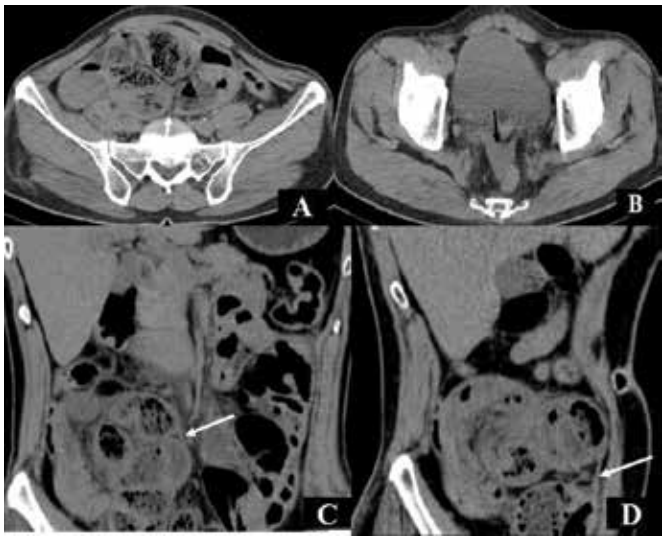

Fig. 2: CT scan showing a conglomerate of multiple intestinal loops surrounded by a thick sac-like membrane (white arrow), dilated small bowel loops and a small amount of ascitic fluid (black arrow) (A and B: axial view; C: coronal view; D: sagittal view).

The patient was hospitalized for emergency exploratory laparotomy. His intraoperative findings showed the encapsulation of the small bowel by a cocoon-like fibrous membrane (Fig. 3), and intestinal wall oedema was seen in the terminal ileum. The lysis of the membrane was carried out, his loops were separated by dissection and the adhesive loops of his small bowel were freed. His freed small bowel segments were viable (Fig. 3). His other abdominal organs were normal.

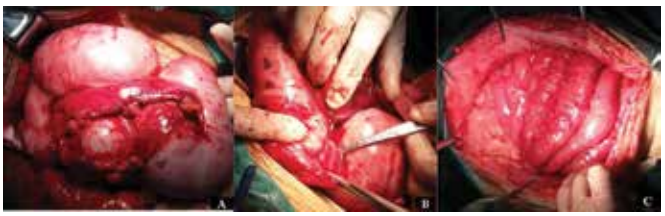

Fig. 3: Operative views: (A) showing the 'cocoon' coverage of the small bowel. (B) The cocoon was opened to release the contained small bowel loops, and the adherent loops of the small bowel were freed. (C) Complete excision of the sac and the release of the entire small bowel.

The histological examination of the membrane revealed fibrous tissue focally lined by flattened mesothelial cells with scattered mononuclear inflammatory cell infiltrate and tissue culture fibroblasts. The patient recovered well and without developing any recurrence at the follow-up 4 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 the 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 aetiology is still uncertain. It could be primary or secondary. The primary condition is idiopathic, and the secondary condition is commonly seen in conditions that result in chronic asymptomatic peritonitis (such as the use of practolol, peritoneal dialysis, endometriosis and abdominal tuberculosis). Moreover, the 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 might also play a role in the aetiology. Even so, more work has to be done to elucidate the exact aetiology 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 non-specific and vary between patients. Despite these difficulties, four main clinical features were 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, the presentation with symptoms suggestive of bowel obstruction but absence of cardinal symptoms such as distension; fourth, the presence of soft non-tender abdominal mass. Our case fits most of the above-mentioned features except for the 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 the preoperative diagnosis of the disease, as shown in our case report. The abdominal radiography hinted a provisional diagnosis of intestinal obstruction. The multi-detector row computed tomography (CT), especially the contrast-enhanced CT, showed that clusters of intestinal loops were enclosed within a membrane-like sac, bowel wall thickening and localized fluid collections. The ultrasound presented a thick-walled mass containing dilated and clumped bowel loops. These imaging features suggest the diagnosis of abdominal cocoon. Explorative laparotomy and laparoscopy may improve the diagnostic accuracy when the membrane encased bowel loops are seen. Some studies had shown that the cocoon usually involved varying parts of the small intestine, but can extend on to cover the colon, the stomach, the liver and even the spleen (9).

Surgery is the treatment method of choice for symptomatic abdominal cocoon in almost all the 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. The intraoperative findings of our patient showed coils of his intestine covered with thick fibrous membrane and the adhesions between the coils. The resection of the encapsulating membrane will return bowel loops to his peritoneal cavity. The breaking of the adhesions needs to be done carefully to prevent damage to the serosal surface and perforation. In our case, the encapsulating membrane was excised and the adhesions were released. The resection of his bowel was not required because his 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 its preoperative diagnosis being difficult. The combination of the patient's physical examination and imaging features, and the knowledge of his accurate medical history, might be helpful in his 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 the patient's prognosis in this rare condition (10).

AUTHORS' NOTE

YS initiated the manuscript, prepared draft of the manuscript. YS, HC, RT, LS, XL, and XZ did the literature review and participated in the discussion of the cases. ZS revised and approved the final draft. The patient's consent was obtained and is available for review at the editor's request.

REFERENCES

1. Jayant M, Kaushik R. Abdominal cocoon in a young man. World J Emerg Med 2014; **5:** 234–6.
2. Yeniay L, Karaca CA, Calışkan C, Fırat O, Ersin SM, Akgün E. Abdominal cocoon syndrome as a rare cause of mechanical bowel obstruction: report of two cases. Ulus Travma Acil Cerrahi Derg 2011; **17:** 557–60.
3. Owtschinnikow PJ. Peritonitis chronica fibrosa incapsulata. Archiv fur Klinische Chirurgie 1907; **83:** 623–34.
4. Foo KT, Ng KC, Rauff A, Foong WC, Sinniah R. Unusual small intestinal obstruction in girls: the abdominal cocoon. Br J Surg 1978; **65:** 427–30.
5. Usha M, Kumar V, Rau Aarathi R, Kamath S. Perforated GIST in jejunum—a rare cause of abdominal cocoon. J Clin Diagn Res 2014; **8:** 132–3.
6. Caldwell J, Dyer A. Sclerosing encapsulating peritonitis (cocoon bowel) presenting after laparotomy for splenic abscess. J Radiol Case Rep 2013; **7:** 17–23.
7. Uzunoglu Y, Altintoprak F, Yalkin O, Gunduz Y, Cakmak G, Ozkan OV et al. Rare etiology of mechanical intestinal obstruction: abdominal cocoon syndrome. World J Clin Cases 2014; **2:** 728–31.
8. Yip FW, Lee SH. The abdominal cocoon. Aust N Z J Surg 1992; **62:** 638–42.
9. Hu D, Wang R, Xiong T, Zhang HW. Successful delivery after IVF-ET in an abdominal cocoon patient: case report and literature review. Int J Clin Exp Pathol 2013; **6:** 994–7.
10. She HL, Ip PP, Cheung SC. Abdominal cocoon: uncommon cause of intestinal obstruction in peritoneal dialysis patient. Hong Kong Med J 2012; **18:** 539.e1–2.