

Diagnosis and Treatment of 26 Cases of Abdominal Cocoon

Sheng Li^{1,2,3} · Jun-Jiang Wang^{1,3} · Wei-xian Hu^{1,3} · Mou-Cheng Zhang^{1,3,4} ·
Xian-Yan Liu² · Yong Li³ · Guan-Fu Cai³ · Sen-Lin Liu² · Xue-Qing Yao^{1,3}

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Abstract

Background and aims Abdominal cocoon (AC) is a rare abdominal disease with nonspecific clinical features, and it is difficult to be diagnosed before operation and hard to be treated in clinical practice. The aim of this study is to investigate the diagnosis and treatment of AC.

Methods The clinical manifestations, findings during surgery, treatments, and follow-up results of 26 cases of AC were retrospectively studied from January 2001 to January 2015.

Results All of 26 cases were diagnosed as AC definitely by laparotomy or laparoscopic surgery. Their clinical findings were various, with 7 intestines obstructed with bezoars and 4 intestines perforated by spiny material. Based on the existence of the second enterocoelia, all cases were categorized into 2 types: type I is absent of second enterocoelia (18 cases, 69.23%), while type II shows second enterocoelia (8 cases, 30.77%). Twenty cases (12 were type I and 8 were type II) underwent membrane excision and careful enterodialysis to release the small intestine entirely or partially, while the other 6 cases (all were type I) did not. In addition, all patients were treated with medical treatment and healthy diet and lifestyle. Finally, most of the patients recovered smoothly.

Conclusions AC can be categorized into two types; surgery is recommended for type II and part of type I with severe complications, but sometimes conservative therapy might be appropriate for type I. Laparoscopic surgery plays an important role in the diagnosis and treatment of AC. Furthermore, favorite health education, healthy diet and lifestyle are of significance in patients' recovery.

Sheng Li, Jun-Jiang Wang, Wei-xian Hu and Mou-Cheng Zhang have contributed equally to this study and share first authorship.

✉ Xue-Qing Yao
yjb9211@21cn.com

- ¹ Southern Medical University, Guangzhou, China
- ² Department of Gastrointestinal Surgery, Central Hospital of Shaoyang, Shaoyang, Hunan, China
- ³ Department of General Surgery, Guangdong General Hospital and Guangdong Academy of Medical Sciences, Guangzhou, China
- ⁴ Department of Gastrointestinal Surgery, First Hospital of Ningbo, Ningbo, Zhejiang, China

Introduction

Abdominal cocoon (AC), also known as “idiopathic encapsulating peritoneal sclerosis (IEPS),” first described by Foo et al. [1] is a rare disease characterized by total or partial encasement of the small bowel by a thick (or thin) fibrous membrane [2]. For its clinical features are non-specific, preoperative diagnosis is difficult. The exact etiology and pathogenesis are still unclear, and its treatment remains controversial. In the present article, we collected 26 cases of AC from January 2001 to January 2015. The clinical manifestations, diagnoses, treatments, and follow-up results were analyzed to investigate the diagnosis and treatment of AC.

Methods

Data review

The case files of AC were extracted from the medical record library of the 3 hospitals (the Central Hospital of Shaoyang, the Guangdong General Hospital and the First Hospital of Ningbo) from January 2001 to January 2015. Data including clinical manifestations, auxiliary examinations, diagnoses, findings during surgery, and treatments of the 26 AC patients were reviewed in detail.

Follow-up

All patients were followed up to investigate the prognosis. A standardized questionnaire concerning the post-discharge medical records, especially the abdominal surgical records, and alimentary symptoms (e.g., abdominal pain, abdominal distention, nausea and vomiting) was conducted by telephone or outpatient service.

Results

Clinical data

A total of 26 cases [14 males and 12 females patients with a mean age of 42.6 years (range 13–78)] were diagnosed as AC, of which 5 cases were combined with cryptorchidism (5/14) and 3 with infertility (3/12), respectively. Sixteen cases (61.54%) were hospitalized with complete or incomplete intestinal obstruction, 7(26.92%) with unexplained abdominal pain, and 3(11.54%) with appendicitis. All were without the history of abdominal trauma or surgery, or long time of oral medication intake, peritoneal dialysis, or intraperitoneal chemotherapy.

All patients (excluding 3 cases of appendicitis) underwent abdominal computed tomography examination, whose results showed partial small intestinal dilation (Fig. 1), and sometimes air fluid levels could be seen; in particular, in 5 cases the membranes that encapsulated the dilated small bowel were shown (Fig. 2). Since abdominal cocoon is a rare disease without specific clinical features, preoperative diagnoses were appropriately achieved in only 3 cases.

Findings during surgery

All of 26 cases were intraoperatively diagnosed as AC by laparotomy (14, 54%) or laparoscopic surgery (12, 46%) (Figs. 3, 4, 5). In all the patients, we found that partial or the entire small bowel was encapsulated in a gray-white,



Fig. 1 CT manifestation of type I: part of intestine expanded slightly, and the fiber wrap could not be seen

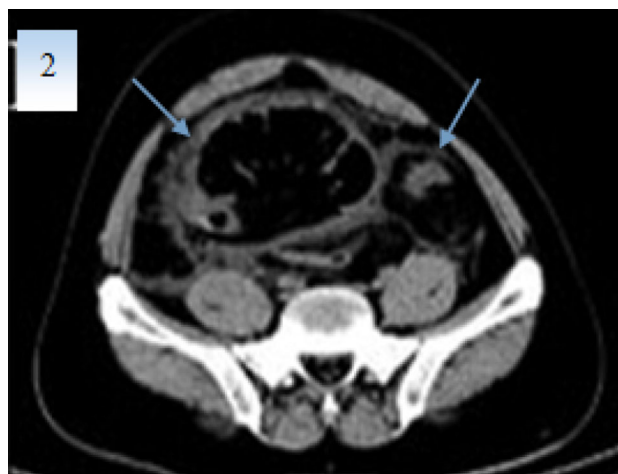


Fig. 2 CT manifestation of type II: the fiber wraps could be seen clearly

fibrous, cocoon-like membrane. In 10 cases (38.46%), the absent of greater omentum and in 6 cases (23.08%), the greater omentum dysplasia had been found. In 7 cases (26.92%), the intestine was obstructed with bezoars. In 4 cases (15.38%), the intestine was perforated by spiny material like bony spur or date pit, including one case which was hospitalized as appendicitis, but the appendix was found normal and the terminal ileum was perforated by a date pit. Appendiceal fecaliths were found in 6 cases (23.08%), and one case was combined with ischemic bowel necrosis. Based on the existence of the second enterocoele, all cases were categorized into 2 types: type I is absent of second enterocoele (18 cases, 69.23%), while type II

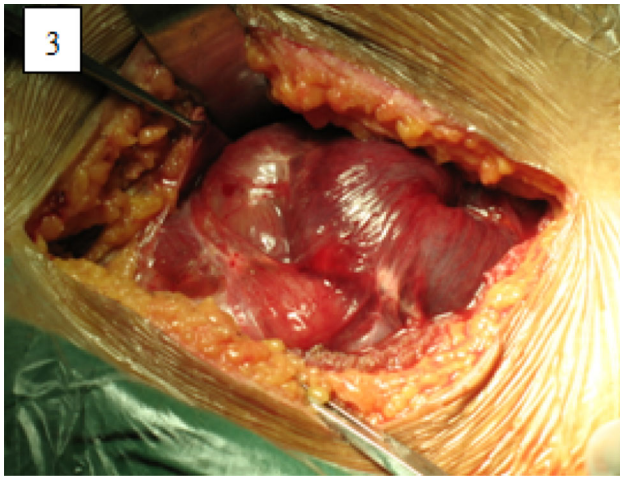


Fig. 3 Type I: small intestine was wrapped by fiber membrane, and the intestine could be seen under the membrane vaguely

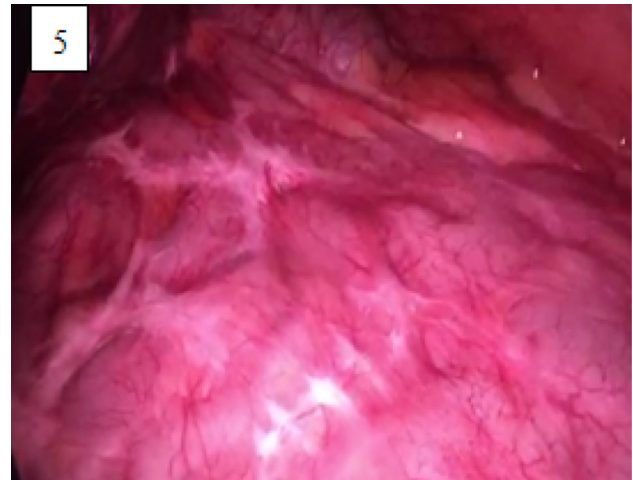


Fig. 5 In laparoscopic view, the total small bowel was wrapped in a fibrous membrane. And the intestinal canal could be seen under the membrane vaguely, with thickened fiber rope belt, and the greater omentum was absent too

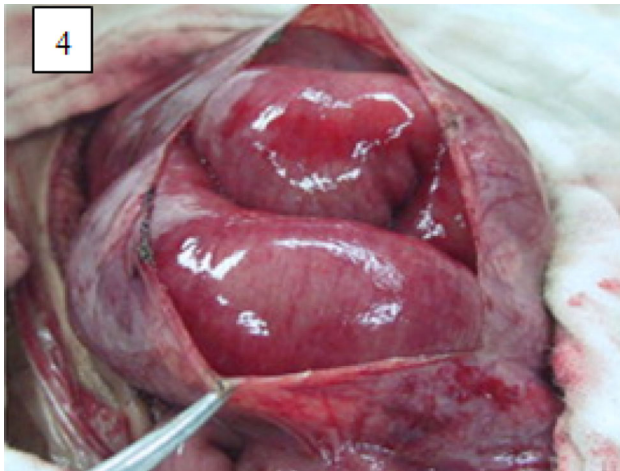


Fig. 4 Type II: the fiber membrane wrapped the intestine to form a second enterocoele

shows second enterocoele (8 cases, 30.77%) (Figs. 3, 4) [3] (Table 1).

Treatments

In 16 cases with intestinal obstruction, 10 cases underwent encapsulated membrane excision and intestinal adhesion released entirely and 6 cases partially (Fig. 6). In 7 cases with unexplained abdominal pain, one case was excised the membranes entirely and 2 partially, while in the other 4 cases, the operations were terminated without any further surgery after the diagnosis of AC by laparoscopy since intestinal dilation was not severe, encapsulated membrane excision was not performed to minimize damage. Preoperative appendicitis diagnosis was made in 3 cases: 2 cases

just underwent appendectomy, and the other one underwent entire membranes excision; intestinal adhesion lysis due to the appendix was almost normal, the terminal ileum was perforated by a date pit, then the date pit was removed, and perforation was repaired. During the operations, 6 cases with appendiceal fecaliths underwent appendectomy besides the 2 appendix cases. The 4 cases with intestine perforated by spiny material underwent foreign material extraction and intestine mending surgery. The case with ischemic bowel necrosis underwent necrotic intestine resection (Table 1).

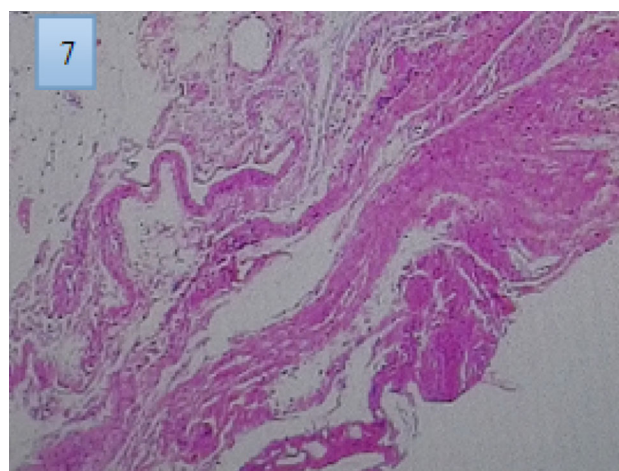
All membrane tissues were examined by optical microscope, and proliferation of fibrocytes and enrichment of collagen fiber were demonstrated, with nonspecific inflammatory reaction and vascular proliferation in some cases (Fig. 7).

All patients were treated with appropriate perioperative treatment, such as parenteral nutrition, anti-inflammatory, and somatostatin based on the patient's condition. All patients were guided with healthy diet and lifestyle (e.g., light diet, eating smaller and more frequently; avoiding raw or cold food, fat, acrimony etc stodgy reach have stronger excitant food, especially avoiding eating too much persimmons and hawthorns for they are easy to form bezoars in human's gastrointestinal tract; and avoiding swallowing bony spur or date pit, etc., which may pierce the intestinal; appropriate physical exercise and avoiding strenuous exercise immediately after a hefty meal for it may cause acute volvulus; keeping the abdomen warm).

Most of the patients recovered smoothly, and the average length of hospitalization was 12.5 days (ranged from 8

Table 1 Clinical data of 26 cases of abdominal cocoon

Clinical data	No. of cases	Ratio (%)
Gender		
Male	14	53.85
Female	12	46.15
Pre-hospital diagnosis		
Intestinal obstruction	16	61.54
Unexplained abdominal pain	7	26.92
Appendicitis	3	11.54
Diagnostic approaches		
Preoperative diagnosis	3	11.54
Diagnosis during operation	23	88.46
Laparotomy	14	54
Laparoscopic surgery	12	46
Findings during surgery		
Small intestine encapsulated in a “cocoon-like” capsule	26	100.0
Lack of greater omentum	10	38.46
Greater omentum dysplasia	6	23.08
Obstruction with bezoars	7	26.92
Intestinal perforation by spiny material like bony spur or date pit	4	15.38
Intestinal perforation with date pit misdiagnosed as appendicitis	1	3.85
Appendiceal fecaliths	6	23.08
Appendicitis	2	7.69
Ischemic bowel necrosis	1	3.85
Clinical classification		
Type I	18	69.23
Type II	8	30.77
The processing means of encapsulated membranes and adhesion		
Entirely excised and released	12	46.15
Partially excised and released	8	30.77
Not treated	6	23.08

**Fig. 6** Encapsulated membranes were excised, and intestinal adhesion was released**Fig. 7** Histologic examination of the membrane tissues shows proliferation of fibrocytes and enrichment of collagen fiber with nonspecific inflammatory reaction

to 21 days). Four cases presented early postoperative inflammatory bowel obstruction (EPISBO) and were cured by hormone therapy (e.g., dexamethasone 5 mg iv Q8h 3–5 days) with nutritional support and administration of somatostatin [4]. There was no perioperative mortality in our cohort.

All patients were followed up [mean 56 months (ranged from 12 months to 10 years)]. Adhesive ileus occurred in 4 patients (15.38%) between 3 and 15 months after surgery, 3 cases recovered by conservative treatment (e.g., sufficient drainage, nutritional support, and administration of somatostatin), and the other one who underwent membranes excision was operated again. During the operation, we found his intestine was adhered again. The other patients were in satisfactory condition with little indisposition (e.g., abdominal pain, abdominal distention, nausea, and vomiting). There is no statistical significant evidence to show any correlation between partial excision and no excision with post-op ileus. In these 4 patients with post-op ileus, 2 of them underwent total excision, 1 partial excision, and 1 no excision, and we believe the post-op ileus was caused by adhesion, which is a common complication of any abdominal surgery.

Discussion

Abdominal cocoon (AC), also known as “idiopathic encapsulating peritoneal sclerosis” [5, 6], is a rare disease characterized by a thick or slender, fibrous membrane that partially or entirely encases the small bowel and that can extend to involve other organs, and it mainly presents in female in tropical and subtropical countries [7, 8], but our data show that male cases are slightly more common than female cases. Much more samples are still required to investigate whether there is a gender difference in AC. Also, the regional disparity requires further research.

Etiology and pathogenesis

The exactly etiology and pathogenesis of AC remain unknown, and many hypotheses have been proposed [5, 8]. Some of these hypotheses involve retrograde menstruation with a superimposed viral infection, retrograde peritonitis via the fallopian tubes, and cell-mediated immunological tissue damage secondary to gynecological infection [5, 9–13]. But it cannot account for the fact that AC also develops in men, premenopausal women, and children.

Since the greater omentum is always dysplastic in almost of AC [3, 14], our viewpoint is much more inclined to the theory that AC is regarded as the result of omental congenital dysplasia [3, 5, 15, 16]. The membranaceous dysplastic omentum encases or adheres to the intestines

and other abdominal organs (e.g., ascending colon, ovary, fallopian tube, testicle), which lead to the dysfunction of organs and present some symptoms. As the result of the obstruction and constraint of membrane, the intestines are unable to move smoothly, especially after heavy food; therefore, the symptom of abdominal pain and (or) ileus would take place, the ovum cannot enter the fallopian tube to be fertilized, and infertility would present; in male patients, the testicle cannot descend into the scrotum at infant stage may cause cryptorchidism [14]. In our data, 16 cases combined with dysplastic (or lack of) omentum (61.54%), 5 with cryptorchidism and 3 with infertility, which supports the hypotheses. We believe the encapsulation of ovary is one of the reasons for infertility, as one of the infertile patients was pregnant two years after the surgery, while the fertility of the other two is unclear due to loss of the follow-up.

Clinical presentation

Abdominal cocoon may have been existed in the patients for a long time before it is diagnosed by incident. Its clinical signs and symptoms vary with the severity and duration of the disease, underlying causes, and the patient's immunological status. We believe there is a close relationship between the symptoms of AC and the variation of life style: small intestinal peristalsis increases severely when abdomen is stimulated by cold, or when greasy, heavy food is ingested. The fibrous membrane covering small intestine in AC is unable to dilate vigorously; as a result, symptoms such as obstruction and abdominal pain are presented. AC most commonly manifests as recurrent acute, subacute, or chronic episodes of intestinal obstruction [12, 17], and abdominal mass can be palpated in some patients. However, some cases may also manifest with more uncommon, but life-threatening complications, including enterobrosis, intestinal necrosis, and malnutrition. Our data showed 7 cases (26.92%) of ileus were caused by bezoars, and 4 cases (15.38%) of enterobrosis were caused by swallowing bony spur or date pit by accident. Therefore, it is significant to cultivate a healthy diet and lifestyle for patients who suspected to suffer from AC.

Diagnosis and classification

It is crucial to differentiate abdominal cocoon from secondary Sclerosing Encapsulating Peritonitis (SSEP) [2]. SSEP is also characterized by a thick, fibrous membrane that encases the small bowel and other organs, but it always has definitive history of long-term peritoneal dialysis, or history of abdominal trauma or surgery, or long time of oral drugs, etc. [8, 18].

Since the characteristic clinical presentation is absent, the preoperative diagnosis of AC is usually difficult. In our data, there were only 3 cases diagnosed before operation. The examination of barium-contrast X-ray and high-resolution magnetic resonance imaging (MRI) or computed tomography (CT) might be helpful for the definitive preoperative diagnosis of AC to some extent [19–23].

However, barium-contrast X-ray should be cautious for it may aggravate the obstructive symptoms of patients with ileus. Accuracy of diagnosis by CT or MRI is limited by its resolution and the physician's experiences. As the result of the low resolution of CT and physicians with insufficient recognition of AC, AC had always been misdiagnosed as intestinal obstruction.

Although recent advances in CT devices that allow multiplanar imaging have enabled preoperative diagnosis of AC, most cases are still incidentally diagnosed during laparotomy. Laparoscopy plays an important role as it can be used for both diagnostic and therapeutic purposes in patient with unclear diagnosis [24–26]. The technique can provide us more accurate information on the degree of obstruction and the types of bowel loops involved; membrane excision and adhesiolysis can also be done with minimal invasion under laparoscopy; and laparotomy can be switched at any time when difficulties were encountered.

Based on the extent of the encasing membrane, AC is classified into 3 types from the literature review [3, 5]: type I—the membrane encapsulated the partial intestine; type II—the entire intestine was encapsulated by the membrane; and type III—the entire intestine and other organs were encapsulated by the membrane. This classification is helpful for the description of AC, but it may not be useful for guiding clinical treatment and management. Based on the existence of the second enterocoelia, we categorize AC into two types and their symptom, sign, CT magnification, the thickness of membrane, and relationship between bowels and membrane are different (Table 2): type I—the

fiber membrane is always slender, the intestine could be seen under the membrane vaguely without the second enterocoelia (Figs. 1, 3), its main clinical presentation may be unexplained abdominal pain or intestinal obstruction, few with abdominal mass, and the membrane always cannot be seen in CT; type II—the fiber membrane is thick and can be shown from high-resolution MRI or CT, the second enterocoelia is formed (Figs. 2, 4), and its main clinical presentation may be intestinal obstruction, always with abdominal mass.

Moreover, their therapies may also be different. Since postoperative adhesion is inevitable, complications such as adhesions will increase with the frequency of operation [27, 28] and type I with asymptomatic or minimal symptoms can be treated with conservative method [2]. However, because of the obstruction of membrane, the bowel in the second enterocoelia is encased and cannot move or expand smoothly, ileus and other unpleasant symptoms would come up sooner or later, and therefore it is recommended to dissect the membrane to release the intestines in Type II.

In our cohort, there were 18 cases of type I (69.23%), 8 of type II (30.77%), and 3 cases diagnosed preoperatively of type II. Whether this kind of classification is appropriate and whether there is a significant difference between the two types still require a larger sample set and further studies to prove (Table 3).

Treatment and management

Surgery remains the gold-standard treatment for AC [3, 29]. However, the operation time is always long and difficult, and it is susceptible to recurrence from intestinal adhesion and obstruction after surgery. Therefore, the treatment should be varied with each individual. Those

Table 2 Classification of abdominal cocoon

	Type I	Type II
Second enterocoelia	No	Yes
Main clinical presentation	Unexplained abdominal pain	Intestinal obstruction
Abdominal mass	Few	Always
Thickness of membrane	Slender	Thick
Membrane can be seen in CT	Few	Always
Need of membrane excision	Few	Always
Conservative treatment	Always	Few

Table 3 Classification of 26 abdominal cocoons

	Type I		Type II	
	n	n/18	n	n/8
Second enterocoelia	0	0	8	100%
Main clinical presentations				
Unexplained abdominal pain	10	55.56%	2	25%
Intestinal obstruction	8	44.44%	6	75%
Abdominal mass	0	0	5	62%
Membrane can be seen in CT	2	11.11%	6	75%
The processing means of encapsulated membranes and adhesion				
Entirely excised and released	8	44.44%	4	50%
Partially excised and released	4	22.22%	4	50%
Not treated	6	33.33%	0	0
Total of cases	18	100%	8	100%

patients discovered by incident with asymptomatic or minimal symptomatic (especial type I) can be managed with conservative treatment (e.g., bowel rest, nutritional support, anti-inflammatory and somatostatin up to the patient's condition), and healthy diet and lifestyle may provide successful outcomes. In our data, there were 8 cases belonged to this condition and recovered well. For those patients of ileus with obvious abdominal pains, distension, and peritonitis, it is suggested to undergo further operation. During surgery, careful dissection and excision of the membrane to release the small intestine is important. It is suggested to excise the appendix filled with stercolith during operation as it would be inflamed in a short time. It is also recommended to examine all the intestines if the condition of appendix is not account for the symptoms in case of missed diagnosis.

Young patient with AC usually has no (or little) uncomfortable symptoms and maybe develop the disease, and the onset is always after inappropriate diet or lifestyle (e.g., eat too much, too heavy, or swallow foreign bodies by accident, catch a cold). In our data, less than half of cases (7 ileuses with bezoars, 4 enterobrosises with swallowed spiny material by accident) were caused by ingesting hard foreign body in meal. Therefore, it is important to educate the patients to develop a healthy diet and lifestyle.

In conclusions, AC is a rare abdominal disease, difficult to be diagnosed preoperatively. Its etiology and pathogenesis remain unclear, and its clinical presentation is nonspecific, when a patient with unexplained pain, abdominal mass, intestinal obstruction, combining with cryptorchid or infertility is encountered; AC should be taken into account. Laparoscopy plays an important role in its diagnosis and therapy. Based on our own experiences, AC could be categorized into 2 types, and this kind of classification may be of great significance for clinical treatment: surgery is recommended for Type II and part of type I with severe complications, but sometimes conservative methods might be appropriate for type I. The aim of operation is to release obstruction but not to dissect all the membrane. To avoid acute onset of AC in asymptomatic patients, education of developing a healthy diet and lifestyle is significant.

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