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ORIGINAL SCIENTIFIC REPORT

Surgical Outcomes in Patients with Abdominal Cocoon: Series of 15 Patients

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Abstract

Background Abdominal cocoon (AC) or sclerosing encapsulating peritonitis is an uncommon cause of intestinal obstruction. Surgical intervention is warranted in patients with persistent pain or intestinal obstruction.

Methodology A retrospective analysis of patients operated for AC was performed. Clinical presentation, radiological data, postoperative outcomes (Ryles tube (RT) removal, duration of hospital stay, enterocutaneous fistula, requirement for re-exploration and mortality) were retrieved and analyzed.

Results Fifteen patients of abdominal cocoon required surgical intervention for various indications. The mean age was 34.46 years (13–60), and 11 (73.3%) were males. Intermittent abdominal pain was present in 14 (93.3%) followed by recurrent subacute intestinal obstruction (SAIO) in 11 (73.3%). Three patients presented with intestinal perforation. Of the 14 patients with preoperative computed tomography, radiological diagnosis was possible in five patients. The mean duration for surgery was 159 min (60–360 min). Membrane encasement was complete in 9/15 and partial in 6/15 patients. Adhesiolysis was done in all patients (complete—10/15 and partial—5/15). Mean duration for RT removal and hospital stay was 4.3 and 12.3 days, respectively. Recurrence of SAIO was observed in three patients, and one patient needed re-exploration for the same. One patient developed postoperative enterocutaneous fistula requiring surgical intervention. Overall mortality in the study was 13.3% (2/15). Four patients had underlying tuberculosis, and the rest were idiopathic.

Conclusion Etiology of AC is not known in majority of patients. Persistent pain and recurrent SAIO are the most common indications for surgery. This morbidity associated with surgery can be reduced by meticulous dissection techniques and appropriate peri-operative care.

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Introduction

Abdominal cocoon (AC) or sclerosing encapsulating peritonitis (SEP) is a disorder characterized by the encasement of small bowel, and sometimes also of the large bowel, by a fibrocollagenous cocoon-like sac. Abdominal cocoon could be primary (idiopathic) or secondary. Secondary form of abdominal cocoon has been reported following a wide range of causes including tuberculosis and post-peritoneal dialysis [1].

The spectrum of presentation ranges from incidental diagnosis in asymptomatic individuals to abdominal pain and acute or recurrent subacute intestinal obstruction [2]. Management of this condition includes treatment of the primary cause if identified. Role of steroids and tamoxifen in management of some patients has been suggested [3]. In patients who do not improve with conservative management or those with recurred episodes of intestinal obstruction or complications such as perforation warrant surgical intervention.

The literature on outcomes of surgery in symptomatic abdominal cocoon is sparse, and surgical outcomes have been reported to be poor [4]. Many patients of abdominal cocoon are managed conservatively; however, the option for surgery is inevitable in patients who fail to improve symptomatically on conservative management or in patients who present with acute intestinal obstruction or features suggestive of perforation peritonitis. Surgical management may include adhesiolysis, small bowel plication, resection of pathological bowel or combination of these procedures. High risk of inadvertent enterotomies exists given the dense and fibrotic inter-bowel adhesions and poor nutritional status of the patient, and could lead to enterocutaneous fistulae. Moreover, risk of postoperative intestinal obstruction also exists [5]. We report our experience of surgery in symptomatic abdominal cocoon patients from a high volume tertiary care center in the northern part of India. In this study, we intend to describe the difficulties in preoperative diagnosis, intra-operative surgical difficulties and postoperative complications and outcomes in abdominal cocoon.

Methods

Study design and setting

This study is a retrospective analysis of prospectively maintained database for abdominal cocoon operated in the department of general surgery from April 2016–August 2018 from a large tertiary care institution which serves as a referral center for North India. The case files were also retrieved for missing information. The details of clinical presentation, underlying etiology, preoperative evaluation and workup, indication for surgery, operative details, postoperative complications, outcomes of surgery and follow-up of these patients were recorded and analyzed.

Criteria for diagnosis

The diagnosis of abdominal cocoon was established on the basis of the presence of a membrane around small bowel loops with or without involvement of large bowel or other organs [6]. Only patients with confirmed intra-operative diagnosis of abdominal cocoon were included in the study irrespective of the indication for the surgery. The underlying etiology was diagnosed on basis of the presence of predisposing factors such as drug history and other associated operative findings. Tuberculosis was considered as the underlying etiology of abdominal cocoon in patients, either based on consistent intra-operative findings and histology or a prior diagnosis based on various parameters such as ascitic fluid analysis, cytology from associated lymph nodes/omental/ peritoneal thickening, etc., who were on antitubercular therapy [7].

Preoperative evaluation

All patients underwent base line investigations, that includes complete hemogram, renal function test, liver function test and chest radiograph. The previous or current history of tuberculosis, drug intake, HIV, and previous surgery were recorded.

Surgery and outcomes

All patients underwent exploratory laparotomy, and surgical findings were noted. Adhesiolysis was attempted in all the patients very carefully to prevent any inadvertent injury to bowel. The membranes and inter-loop adhesion were resected with scissors. Postoperative outcomes that include duration of Ryles tube (RT) requirement, postoperative complications, total in-hospital stay, need for re-exploration and mortality were recorded. Follow-up of the patients was recorded in outpatient clinic or through telephone.

Results

Clinical presentation

A total of 15 patients were included in this study. The mean age of the study population was 34.46 (range 13–60 years) with male preponderance ($n - 11$). The most commonly

presenting symptom was abdominal pain (93.3%). The most common indication of laparotomy was intestinal obstruction (80%) followed by perforation peritonitis (20%). Seventy-three percent of patients gave history of recurrent transient episodes of abdominal distension and discomfort which relieved spontaneously before the index presentation. The clinical profile is shown in Table 1.

Four out of 15 patients (26.6%) were diagnosed with tubercular abdominal cocoon. Two out of these four patients were diagnosed with cases of abdominal TB based on high adenosine deaminase (ADA) level from peritoneal fluid analysis and had already completed the ATT few months before index surgery and the other two patients had evidence of epithelioid granulomas with caseating necrosis suggestive of TB on histopathology who were postoperatively started on ATT and received ATT for 6 months.

The preoperative contrast-enhanced CT (CECT) scan was performed in 14 patients. The preoperative diagnosis of sclerosing encapsulating peritonitis based on CECT abdomen (Fig. 1a) was made in 35.7% patients (5/14). One patient who presented with frank peritonitis underwent surgery without preoperative CECT abdomen.

Operative details and postoperative course

During surgery, compete cocoon encasing the entire small and large intestines was found in 60% patients and partial cocoon encasing only the small bowel loops was identified in 40% patients (Fig. 1b). Two patients had associated small bowel stricture and underwent adhesiolysis and resection of stricture bearing segment stoma formation.

Table 1 Clinical profile of operated patients

Mean age (range) (Y)	34.46 (13–60)
Male/female	11: 4
Clinical presentation (%)	
Abdominal pain	14/15 (93.3%)
Abdominal distension	14/15 (93.3%)
Vomiting	14/15 (93.3%)
Loss of appetite	11/15 (73.3%)
Recurrent intestinal obstruction in the past	08/15 (53.3%)
Clinical findings (%)	
Pallor	05/15 (33.3%)
Generalized lymphadenopathy	Nil
Pedal edema	02/15 (13.3%)
Peritonitis	03/15 (20%)
Ascites	01/15 (6.66%)
Indication for surgery	
Acute intestinal obstruction	12/15 (80%)
Peritonitis	03/15 (20%)

Three patients had small bowel perforation with gross contamination of which one had tuberculosis as the underlying cause. Resection of perforation bearing segment and stoma was made in two patients, and in one patient the bowel could not be exteriorized in view of frozen abdomen. Adhesiolysis was attempted in all the patients. However, complete adhesiolysis could not be achieved in five patients (33.3%) owing to dense adhesions and increased risk of enterotomies. Three of five patients managed with an enterostomy proximal to the level of obstruction based on the surgeons' discretion. A total of seven patients (46.6%) in the study population needed an enterostomy; of these, five were ileostomies and two were jejunostomies. Abdominal closure could be achieved in all the patients except in one patient, whose laparostomy was managed conservatively with wound manager. The operative details are shown in Table 2.

Compete symptomatic improvement could be achieved in 86.6% (13/15) of patients in the immediate postoperative period. The mean duration for the need for nasogastric drainage was 4.3 days, and most patients were started on enteral feeds within 1 week from surgery. The mean in-hospital stay was 12.3 days. Infective complications such as surgical site infections, intra-abdominal collections were seen in four (26.6%) and two (13.3%) patients, respectively, who were managed conservatively with regular aseptic dressings and ultrasound-guided single-time aspirations of abdominal collection. One patient, who underwent adhesiolysis with repair of enterotomy in the index surgery, had an enterocutaneous fistula diagnosed on postoperative day 4. Patient was re-explored, and ileostomy was created from the enterotomy site. Later, the patient had stable postoperative course and was discharged after 10 days. One patient in whom an ileal perforation could not be exteriorized in view of frozen abdomen was managed conservatively with wound manager who later died due to massive pulmonary thromboembolism. One patient (6.6%) who presented with severe peritonitis succumbed to sepsis in immediate postoperative period.

So, overall four patients had superficial surgical site infection, two developed postoperative intra-abdominal collections, one had enterocutaneous fistula requiring early reoperation, and two deaths occurred. The operative outcome details are shown in Table 3.

Follow-up

Thirteen patients discharged were on regular follow-up. Three out of 13 patients (23.07%) had features suggestive of recurrent SAIO over a median follow-up duration of 19 months (range 5–34 months). However, the second surgery for recurrent obstruction was needed in only one patient and the remaining two patients were managed

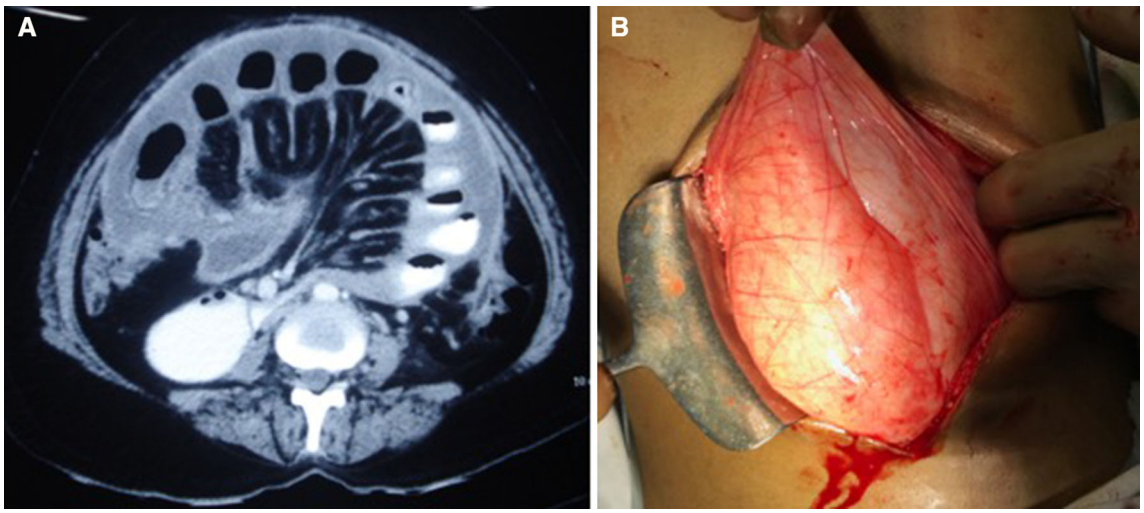

Fig. 1 **a** Contrast-enhanced computed tomography showing membranes encasing the small bowel loops. **b** Intra-operative picture showing complete encasement of bowel loop with cocoon-like membranes

Table 2 Cocoon and associated findings

Type of cocoon	
Complete cocoon	09/15 (60%)
Partial cocoon	6/15 (40%)
Etiology of cocoon	
Non-tubercular	11/15 (73.3%)
Tubercular	04/15 (26.6%)
Adhesiolysis	
Complete	10/15 (66.6%)
Partial	05/15 (33.3%)
Surgery performed	
Adhesiolysis	8/15 (53.3%)
Adhesiolysis with stoma	3/15 (20%)
Resection of stricture and stoma formation	2/15 (13.3%)
Resection of perforation bearing segment with stoma formation	2/15 (13.3%)
Failed adhesiolysis + lavage + B/L pelvic drain	1/15 (6.6%)

conservatively. Stoma reversal was performed in four out of six patients who were discharged with enterostomy, while the remaining two are awaiting surgery.

Discussion

Abdominal cocoon is an uncommon cause of intestinal obstruction, where surgical outcomes are inferior to many other common causes of intestinal obstruction. In our series of 15 patients undergoing surgery for abdominal cocoon, the indications of surgery were intestinal obstruction and intestinal perforation. Complete symptomatic improvement could be achieved in 13 (86.6%) patients in postoperative period. Superficial surgical site infection was the most common postoperative complication (26.6%) followed by infective intra-abdominal collections (13.3%) and entero-cutaneous fistula in one patient. A mortality of 13.3% (2/15) was recorded.

The underlying pathogenesis of abdominal cocoon is poorly understood; and although many theories regarding its causation have been proposed, none is completely accepted [3, 5]. Surgery is an important tool in the management of these patients, especially if the patient does not improve with management of underlying cause and continues to be symptomatic or presents with complications. This, however, comes with the caveat of prolonged duration of surgery, risk of inadvertent enterotomies and morbidity associated with such complications. Conservative management of abdominal cocoon is also shown to be effective, especially in tubercular abdominal cocoon in patients with no features suggestive of acute intestinal obstruction or peritonitis [8, 9]. It is therefore our strategy not to operate asymptomatic individuals if a treatable cause such as tuberculosis is identified and should be treated.

In order to have a detailed preoperative management strategy and to reduce the intra-operative surprises, the need for preoperative diagnosis has achieved a greater emphasis. In various retrospective studies, the definitive preoperative diagnosis could not be achieved in significant number of patients. In a study by Bo Wei et al, only four out of 24 patients could be diagnosed with AC preoperatively by using CT and barium studies [2]. Similarly in another study by Sheng, Li AC was diagnosed preoperatively in only three patients out of 26 [5]. Preoperative

Table 3 Outcome of surgery

Complete adhesiolysis achieved	10/15 (66.6%)
Mean duration of need for Ryles tube placement	4.3 days
Postoperative outcome (immediate)	
Complete symptomatic improvement	13/15 (86.6%)
Need for reoperation	01/15 (6.66%)
Mean in-hospital stay	12.3 days
Postoperative complication	
Infective collections	02/15 (13.3%)
Enterocutaneous fistula	01/15 (6.66%)
Wound site infections	04/15 (26.6%)
Mortality	02/15 (13.3%)
Follow-up	
Recurrent obstruction	03/13 (23.07%)
Need for repeated surgery for obstruction	01/03

diagnosis is difficult because of nonspecific symptomatology and lack of clear-cut radiological criteria for abdominal cocoon. In the present study, preoperative diagnosis could be achieved only in only 35.7% patients only. However, in a recent study by Gorsi et al. various radiological features to diagnose AC on CT abdomen such as cauliflower sign, concertina pattern and the bottle gourd signs were described which improved the sensitivity and specificity of CT, especially in patients with intestinal obstruction at presentation [10]. Cross-sectional imaging has an undeniable role in diagnosing the level of obstruction and to rule out other possible causes for intestinal obstruction [11]. However, most reports suggesting the excellent role of imaging in preoperative diagnosis have done the imaging analysis retrospectively.

Majority of patients who present with abdominal cocoon are nutritionally depleted owing to recurrent subacute intestinal obstructions. However, nutritional optimization is difficult in an acute presentation. Whenever feasible, preoperative nutritional support, preferably enteral nutrition, should be administered. A study by Li [4] showed that preoperative nutritional support marginally reduces the postoperative complication. Surgery in abdominal cocoon is technically challenging and should be performed only by an experienced surgeon. The principle of surgery in abdominal cocoon is compete excision of membrane and adhesiolysis. Manipulation of bowel should be done very gently to avoid inadvertent bowel injury. Dissection and excision of membrane should be preferably done by sharp dissection using scissors. Inappropriate usage of electrocautery should be avoided. Presence of thick fibrotic membranes makes this dissection more cumbersome and increases the risk of bowel injuries. In our series, 66.6% of the patients had thick fibrotic membranes. However, intraoperative enterotomies happened only in one patient.

Surgical intervention in AC carries significant morbidity. Enterocutaneous fistula is the most devastating complication which is mostly caused by the inadvertent bowel injuries that occur during adhesiolysis. ECF further complicates the postoperative course in AC patients who already have poor nutritional status leading to electrolyte disturbances, further deterioration of nutritional status, septic complications, need for reoperation and stoma formation. However, the incidence of ECF following surgery for AC is possibly underreported [4].

In a published case series by Kaushik et al. [12], four out of five patients had postoperative enterocutaneous fistula. However, all these patients could be managed successfully either conservatively or by exteriorization of fistula. Another series of 24 patients reported ECF in only one patient [2]. In the current study, two of our patients developed enterocutaneous fistula postoperatively one from a documented enterotomy in the index surgery and was managed by exteriorization of fistula in a second surgery. Another patient was managed conservatively with wound manager and had eventually succumbed to massive pulmonary thromboembolism during the hospital stay. Incidence of early postoperative small bowel obstruction (EPSBO) following surgery is widely variable. However, majority of these patients improve with conservative management alone. EPSBO is attributed to factors such as inter-bowel conglutination, injury to bowel serosa and inflammatory edema [2]. Meticulous dissection and gentle bowel handling during the adhesiolysis might help in reducing the incidence of EPSBO. Enterostomy as a part of AC surgery definitely adds to the morbidity. However, the incidence of stoma formation is high in the current study (46.6%). The possible reason for this higher rate might be the patient profile with poor nutrition, poor socioeconomic status and delayed presentation to our tertiary care hospital. Various other factors such as need for enterostomy for longer duration, need for repeated surgeries owing to persistent intestinal obstruction, prolonged hospitalization not only increase the morbidity but also increase the costs incurred to the patient.

While our series is a large series on this uncommon condition, it is a retrospective study from a single larger volume center from an area endemic for tuberculosis. Therefore, the findings may not be generalizable to other centers or geographic locations. Still the report adds to the literature on this condition and provides important information for clinicians treating abdominal cocoon.

Conclusion

Etiology of abdominal cocoon is not known in majority of patients. High index of suspicious while evaluating pre-operative cross-sectional imaging improves the diagnostic accuracy. Persistent pain and recurrent SAIO are the most common indications for surgery. However, surgery in abdominal cocoon has increased risk of bowel injury and high postoperative morbidity. This morbidity associated with surgery can be reduced by meticulous dissection techniques and appropriate peri-operative care.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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