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Original Research Article

Primary and secondary abdominal cocoon- diagnostic and management challenges: retrospective study

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ABSTRACT

Background: Abdominal cocoon is a rare cause of intestinal obstruction characterized by fibro collagenous membrane encapsulating the abdominal contents to varying degrees. The most commonly identified etiology remains to be 'idiopathic' and hence it is also known as idiopathic sclerosing encapsulating peritonitis.. Few case reports of abdominal cocoon secondary to gastrointestinal malignancy and tuberculosis have also been reported. The objective of the study was to study the spectrum of clinical presentation, identify the various aetio pathogenesis described and their impact on outcome of surgically managed patients with abdominal cocoon.
Methods: The clinical data of twelve different cases of abdominal cocoon presented to a general surgery unit from January 2012 to December 2017 with minimum of 1 year follow up were analyzed.
Results: In our series we had 12 patients with cocoon who underwent surgical intervention out of which 8 were primary and 4 were secondary to TB. In primary type one out of eight patients had acute presentation, but in secondary three out of four had acute presentation. In primary 6 out of 7 patient's pre-operative CT showed cocoon, but in secondary only 1 out of 3 showed cocoon. In patients with primary cocoon 3 out of 8 patients had bowel resection and only one had post-operative morbidity. In patients with secondary cocoon three patients had bowel resection, stoma creation and reoperations with stormy post operative period. All 4 had post-operative morbidity, however all of them received ATT and definitive surgery was performed 1-2 years later with minimal resection and serial images also showed resolution of cocoon formation with ATT. Primary group up to 5 years follow up, there was no recurrence of symptoms. Both groups did not have any mortality.
Conclusions: Abdominal cocoon is a rare disorder and the cause may be primary or secondary. In our series tuberculosis is the aetiology for secondary cocoon. Primary cocoon is easier to diagnose, manage and associated with less post operative complications compare to secondary cocoon. Damage control first surgery, nutritional build up and treatment with anti tubercular drugs are needed for management of cocoon secondary to TB for a better outcome in acute presentations. Definitive surgery can be performed once the nutritional status improves with less morbidity.

Keywords: Abdominal cocoon, Tubercular abdominal cocoon

INTRODUCTION

The abdominal cocoon is a rare cause of intestinal obstruction. The first case was documented in 1907 by Owtschinnikow, who labelled it peritonitis chronica fibrosa incapsulata.[1] The term abdominal cocoon was later coined by Foo et al. It is classically described as sclerosing encapsulating peritonitis underscoring the characteristic fibrous membrane encapsulating the small bowel.[2] Sclerosing encapsulating peritonitisis classified as primary or secondary based on the etiopathogenesis.[2,3]

Primary sclerosing encapsulating peritonitis or abdominal cocoon syndrome is classified into three types based on

the extent of encasement of abdominal contents by the membrane. Type I and type II have encasement of part or complete intestine by fibrocolagenous membrane respectively. In type 3, small intestine, appendix, caecum, ascending colon, stomach, liver and ovaries are encased.[2]

The primary type is classically described in young adolescent females from tropical and subtropical countries.[4,5] The primary sclerosing encapsulating peritonitis is mostly idiopathic in nature. Few of the etiopathogenesis proposed for primary abdominal cocoon are viral peritonitis, retrograde menstruation with superimposed viral infection and gynaecological infection-inducing cell-mediated immunological tissue damage.[1,2,5,6] However, these theories may not explain the etiopathogenesis in all patients as this condition is also seen in men, premenstrual women and children.[1,2,7] Management of primary abdominal cocoon has evolved over the years ranging from complex procedures like bowel intubation to simple membrane excision.

Secondary abdominal cocoon can occur secondary to multiple triggers like presence of chronic ambulatory peritoneal dialysis, ventriculo-peritoneal and peritoneo venous shunts and is associated with certain medical conditions like SLE, liver cirrhosis, endometriotic cyst, recurrent peritonitis, tuberculosis and malignancy.[2,3,7] Our literature review showed very few case reports of abdominal cocoon secondary to tuberculosis.[8,9]

In a developing country like India, we would expect Tuberculosis to be a major etiology in the non-dialysis group of secondary cocoon.[8,9] Presentation of TB abdomen can vary significantly, and management will depend on severity of presentation. However there was a trend towards non operative approach for abdominal TB emerging.[10] In our retrospective study we had 12 cases of abdominal cocoon which was managed surgically; we have analysed and compared their types, clinical presentations, diagnostic and operative difficulties and outcome.

METHODS

Retrospective analysis of patients diagnosed to have abdominal cocoon and had surgical intervention in a general surgical unit of a tertiary hospital from January 2012 to December 2017 with minimum of 6 months follow up. Out of 12 patients, 8 were primary type and 4 were secondary type. All of them had abdominal pain as common symptom. Many had sub-acute intestinal obstruction as presentation and few presented with peritonitis. All of them underwent some surgical procedure, this varied from simple membrane excision adhesiolysis to complex procedures like stoma creation, bowel resection etc. As described earlier abdominal cocoon can be divided into different types based on extent of involvement. In our study we had 4 patients each in Type 1, 2 and 3 respectively. Operative findings varied from membrane covering part or entire bowel. In secondary type apart from cocoon we also found multiple tubercles, inter loop adhesions and perforations with gross contamination. Postoperative complications also varied from no morbidity to recurrent operation and ICU care. However in our study we had no mortality. Our follow up varied from 6 months to 5 years with no documented recurrence of cocoon formation.

RESULTS

Table 1 describes in detail of above mentioned clinical profile of each patient.

Table 1: Clinical profile of 12 patients with abdominal cocoon.

S. No	Type	Age/ sex	Clinical presentation	Operation done	Operative finding	Post op morbidity	Outcome
1	Primary	43/M	Recurrent abdominal pain for 4 years and abdominal mass for 4 months. No previous operation	Membrane excision and adhesiolysis	150 cm of distal small bowel in the cocoon Type 1	Nil	No recurrence Follow up 5 years
2	Primary	68/M	Recurrent SAIO. K/c/o DM, HTN, HIV positive	Membrane excision and adhesiolysis	Small bowel loops covered in thin membrane. No omentum Type 2	Nil	No recurrence in 1 year follow up
3	Primary	50/M	Recurrent episodes of SAIO- no previous operation	Membrane excision adhesiolysis	Membrane covering small, large bowel, stomach and liver No omentum Type 3	Post op adhesive intestinal obstruction- conservative management	No recurrence in 2 year follow up
4	Primary	41/M	Recurrent episodes of SAIO- no previous operation	Membrane excision adhesiolysis	Membrane covering small and large bowel Type 3	Nil	No recurrence in 3 years follow up

Continued.

S. No	Type	Age/ sex	Clinical presentation	Operation done	Operative finding	Post op morbidity	Outcome
5	Primary	42/M	Abdominal pain and distension	Membrane excision and adhesiolysis	Thin membrane covering stomach to sigmoid Type 3	Post op adhesion with partial obstruction- conservative management	No recurrence in 6 months then last to follow up
6	Primary	44/M	Recurrent episodes of SAIO 2 operations in the past 6 months- no improvement	Membrane excision and adhesiolysis	Membrane covering small bowel, dense adhesion due to previous operation Type 2	Peri- operative nutritional build up with TPN. UTI	No recurrence in 1 year follow up
7	Primary	34/M	Recurrent SAIO – one operation laparoscopic appendicectomy	Membrane excision adhesiolysis and small bowel R&A	Membrane covering small bowel Type 2	Post op adhesion with partial obstruction – conservative management	No recurrence in 1 year follow up
8	Primary	50/F	Abdominal pain, loose stools for 10 days. Peritonitic at presentation	Membrane covering small and large bowel with 2 perforations in caecum and descending colon with peritoneal contamination	Membrane excision peritoneal lavage subtotal colectomy end ileostomy Type 3	Post op ICU stay- recovered well	No recurrence in 6 months follow up
9	Secondary	27/M	Fever, abdominal pain and SAIO- admitted diagnosed as TB with colonoscopy- biopsy. Started ATT. TPN for nutrition. Developed peritonitis	Adhesiolysis proximal stoma distal mucous fistula and peritoneal lavage	Membrane covering small bowel with tubercles in peritoneum, serosa and mesentry. Multiple perforation with gross contamination Type 1	Stormy post op period, ICU care. Bile in drains. Managed conservatively . Recovered with long hospital stay	18 months follow up –no recurrence
10	Secondary	68/F	Abdominal pain and distension for 4 months. K/C/O CAD	Membrane excision adhesiolysis, bladder injury repair	Cocoon covering part of small bowel and tubercles in peritoneum and serosa Type 1	Bladder injury- primary repair	No recurrence in 6 months then last to follow up
11	Secondary	24/F	Abdominal pain, distension low grade fever for 2 years	Laparotomy adhesiolysis proximal stoma and distal mucous fistula peritoneal lavage	Gross peritonea contamination with cocoon covering part of small bowel peritoneal and serosal tubercle. No perforation found Type 1	Reoperation 1 week perforation closure and feeding jejunostomy distal to perforation laparostomy. Stormy post op period recovered with ATT	18 month follow up stoma reversed- no tubercles found in reoperation

Continued.

S. No	Type	Age, sex	Clinical presentation	Operation done	Operative finding	Post op morbidity	Outcome
12	Secondary	33/M	Recurrent SAIO for 1 year. Diagnostic laparoscopy and started ATT. Presented with mass for 10 days	1.Drainage of abscess 2.Laparotomy membrane excision – partial loop ileostomy at perforation site and laparostomy	Collection with enteric content 2.Membrane covering small bowel with tubercles perforation in small bowel Type 2	Damage control surgery then definitive surgery. Recovered well with ATT	12 months follow up no recurrence

Table2: General profiles of patients with primary and secondary abdominal cocoon.

General profile	Primary (N=8)	Secondary (N =4)
Age in years mean (range)	47 (34-69)	38 (27-68)
Sex, M:F	7:1	2:2
Abdominal pain, N (%)	8 (100)	4 (100)
Abdominal distension, N (%)	6 (86)	4 (100)
SAIO, N (%)	5 (71)	4 (100)
Peritonitis, N (%)	1 (12.5)	3 (75)
Hb (mean)	13.1	9.4
Albumin (mean)	3.98	2.55
BMI (Mean)	23.58	20

Table 3: Comparison between primary and secondary cocoon on evaluation, intervention and outcome.

	Primary cocoon (N=8)	Secondary cocoon (N=4)
Peritonitic presentation	1	3
Pre op CT	7	3
Pre op diagnosis of cocoon based on CT	6/7	1/3
Pre op diagnosis of TB	-	3/3
Surgery	8	4
Membrane excision alone	5	1
Damage control surgery	0	3
Bowel resection/stoma	3	3
Confirmation by biopsy	8	4
Morbidity	2	4
Reoperation	0	3
Mortality	0	0

Detail analysis of clinical profile revealed certain differences between primary and secondary type of cocoon which described in Table 2 and 3. Table 2 is showing the comparison of general profile like age, sex, clinical presentation and nutritional status between primary and secondary cocoon patients. In our series, primary cocoon showed a male predominance whereas secondary cocoon had equal distribution. Secondary cocoon also was more likely to present with acute and sub-acute presentation to the healthcare facility. Secondary group had poor nutritional status compared to primary.

Table 3 compared radiological evaluation, operative intervention, operative findings, morbidity and outcome between groups. Primary type 86% had cocoon diagnosis pre operatively. Secondary type only 33% had the diagnosis of cocoon through imaging preoperatively. However TB was diagnosed in 66% of patients with imaging. In secondary group 75% had damage control surgery as compared to none in primary group. Postoperative morbidity was 100% in secondary and 25% in primary group and secondary group had 75% re operation rate compare to none in primary group.

DISCUSSION

Abdominal cocoon can be primary or secondary. Radiologically diagnosed patients with minimal or no symptoms are often managed conservatively.[5,10] Tuberculous abdominal cocoon needs tissue diagnosis before embarking on conservative approach.[10] In primary type chances of recurrent symptoms are higher if managed conservatively.[5] Management of patients who require surgical intervention becomes dicey due to lack of established treatment guidelines.

Clinical presentation of abdominal cocoon may be vague abdominal pain, abdominal mass or they may present with sub-acute symptoms like partial intestinal obstruction or acute presentation like peritonitis. In our series Secondary cocoon had more acute presentations compared to primary group and required emergency operations.[1,7] Clinical examination may be non-specific or patient may have a mass because of clumped up bowel loops. In acute presentations features of peritonitis may be present. Primary cocoon had mild symptoms and less acute presentations compare to secondary group.

Abdominal cocoon used to be an intra-operative surprise for surgeons mostly; however with the availability of cross sectional imaging cocoon can be diagnosed pre operatively.[11] In our study primary type was diagnosed better compare to secondary. In secondary group even

though cocoon was not diagnosed in imaging, tuberculosis was diagnosed in 3 out of 4 cases.

Figure 1: Preoperative CT showing mesenteric fat stranding and clumped up bowel loops in a case of secondary cocoon due to TB.

Figure 2: Postoperative, post ATT, CT showing resolution of mesenteric stranding and clumping of bowel loops of the same patient.

Cross sectional imaging is the investigation of choice for abdominal cocoon. They are useful in diagnosing the cocoon as well as secondary aetiology if present.[11]

Operative procedure for cocoon can be membrane excision, adhesiolysis, bowel intubation or bowel resection.[4] For primary cocoon most of the studies in literature showed that simple procedures like membrane excision and adhesiolysis is adequate as seen in our study.[7,11] Intra operative findings were a dense membrane covering bowel loops forming a lump; however extent of covering was different between cases. But in secondary type due to TB operative procedures were complex like bowel resection, stoma creation, laparostomy etc. Intra operative findings had edematous bowel, caseous tubercles over serosa and peritoneum, enlarged mesenteric nodes, dense inter loop adhesions and multi-level obstructions as described in literature.[8,9,12]

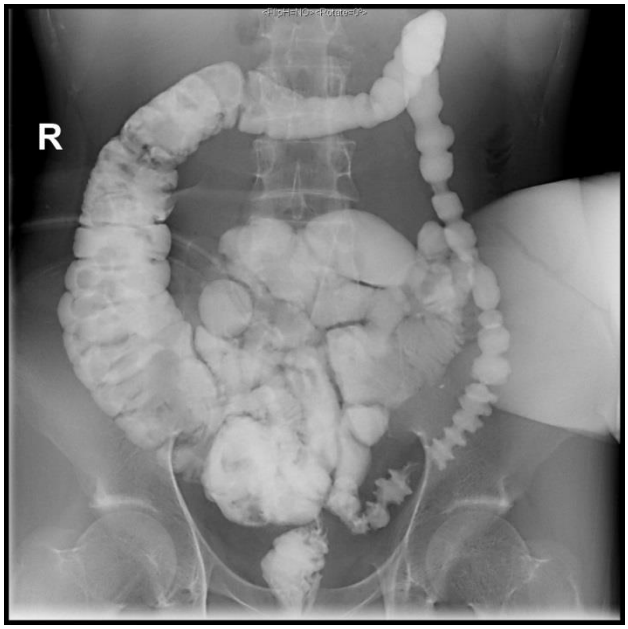

Figure 3: Barium study through mucous fistula showing complete resolution of distal obstruction.

This probably is the reason why secondary cocoon group had complicated surgical procedures and stormy postoperative period compared to primary group. Comparing nutritional parameters like Haemoglobin, Albumin and BMI primary group had a better nutritional status compared to secondary group. One patient in primary group had a poor nutritional status because he was operated elsewhere twice and hospitalised for one month and came to our centre as his symptoms were not resolving. Tuberculosis as a disease process is associated with poor nutritional status and for peritoneal Tb to manifest as cocoon, disease process probably may be going on for a longer duration.[12,13] In primary cocoon operation, an effort is made to release all bowel loops, removal of all membranes and procedure like bowel intubation is done to prevent recurrence, and rarely if required bowel resections was done.[4] When it comes to secondary cocoon due to Tb similar attempts may prove fatal as most patients were in poor nutritional status and present with peritonitis.[14,15] In these situations damage control surgery is done for a proximal diversion, laparostomy and biopsy which will relieve symptoms and provide diagnosis. Once treatment initiated (ATT) and

nutritional build up achieved a definitive surgery can be planned later on.

In our series among 4 cases of secondary cocoon, 3 presented with acute or sub-acute symptoms. An approach of damage control surgery was done for all 3 patients like peritoneal lavage, stoma, mucous fistula creation and laparostomy. Secondary cocoon had histopathological features of tuberculosis in the biopsy. All 3 patients had stormy postoperative period including re operations, ICU stay. However they responded well to ATT treatment and recovered.

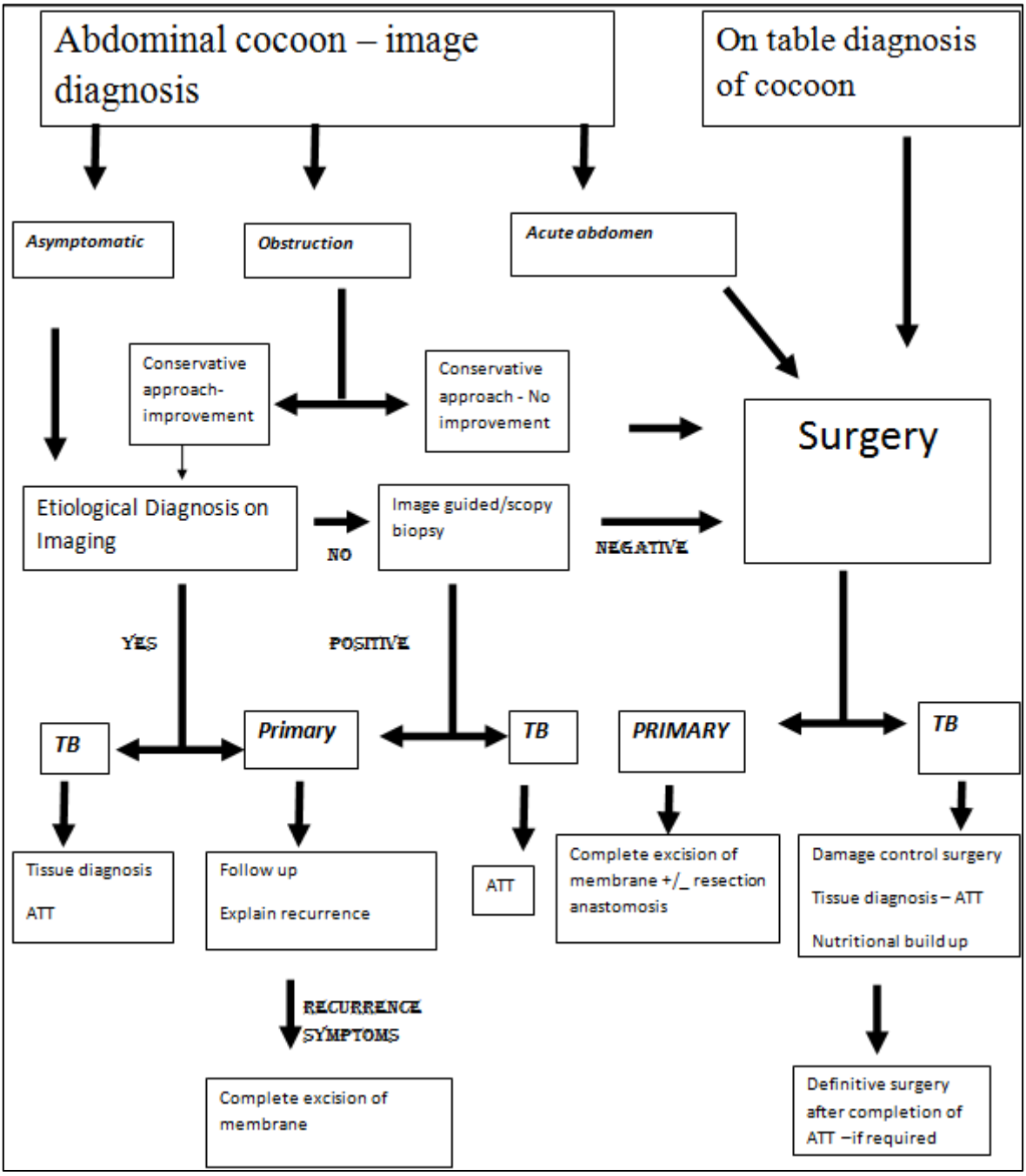

Figure 4: Flow chart for diagnosis and management of abdominal cocoon.

In literature review of abdominal TB, over the years the surgical management of TB has been changing. Before the advent of ATT operative procedures were non curative like bypass surgery. Once ATT came into treatment, the operative procedure aimed at complete eradication of disease like hemi-colectomy etc. Recently due to newer methods of achieving tissue sample for diagnosis and availability of better sensitive and specific tests, operative procedures reduced to diagnostic purpose only.[16] But for abdominal cocoon secondary to TB, there is no estabilished protocol. Tuberculous cocoon with minimal or no symptoms can be managed conservatively with tissue diagnosis followed by anti-tuberculosis treatment.[10] Literature reveals that patients with TB have

poor nutritional status and doing a bowel resection and anastomosis or extensive adhesiolysis will lead to significant postoperative morbidity and mortality.[17] Resecting large segment of bowel will lead to short bowel syndrome and there is enough evidence in literature that with ATT treatment partial obstruction, serosal and peritoneal tubercles can resolve completely or partially. These findings led to the current approach of damage control surgery, ATT treatment followed by definitive surgery as required.[9,12,16] In our study 3 patients had damage control surgery for TB abdominal cocoon. Even though they had a stormy postoperative period, their rehabilitation was good with ATT. Their serial cross sectional imaging revealed reduction in cocoon formation, mesenteric stranding and bowel clumping (Figure 1 and 2). 2 out of 3 underwent stoma reversal without any bowel resection. One awaiting reversal with barium study showing good contrast flow distally (Figure 3).

However in primary type 3/8 patients had bowel resection out of which 1 had stoma and 2 had anastomosis. Only 1/8 patient presented with acute symptoms requiring emergency operation. This is probably because primary cocoon disease process does not involve bowel mucosal surface and nutritional status is better than secondary cocoon due to TB. Here all patients had complete adhesiolysis, membrane excision and bowel resection (if required) in the operation. Primary cocoon histo-pathology revealed deposition of fibrocollagenous tissue in the membrane. Primary cocoon did not have any recurrence in the follow up period of minimum one year and up to 3 years for different patients.

These contrasting findings between primary and secondary cocoon in this study established a need for different management approach for primary cocoon and secondary cocoon due to TB and help us to develop an algorithm. Management algorithm for abdominal cocoon has been described as a flow chart (Figure 4). As seen in the flow chart patients with minimal or no symptoms in both groups can be managed conservatively. However secondary cocoon due to TB needs tissue diagnosis before starting ATT and primary can have worsening or recurrence of symptoms with conservative management. Symptomatic patient needs surgical intervention, primary group needs definitive primary surgery up front but secondary due to TB is best managed with damage control surgery and tissue diagnosis followed by ATT and definitive surgery later if required.

CONCLUSION

Abdominal cocoon is a rare disorder and may be primary or secondary type. In our series tuberculosis is the aetiology for secondary cocoon. Primary cocoon is easier to diagnose, manage and associated with less postoperative complications compare to secondary cocoon. Damage control first surgery, nutritional build up and treatment with anti-tubercular drugs are needed for cocoon secondary to TB for a better outcome in acute presentations. Less damaging definitive surgery can be performed once the nutritional status improves.

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