

## Sclerosing Encapsulating Peritonitis (SEP) due to *Mycobacterium Bovis*

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### ABSTRACT

Sclerosing Encapsulating Peritonitis (SEP), a visceral encapsulation syndrome of inflammatory origin either primary or secondary based on causation, is an infrequent cause of mechanical bowel obstruction that may commonly be seen in young patients. Preoperative diagnosis is quite challenging and usually missed due to the non-specific nature of the clinical picture. In most of the cases, a definitive diagnosis is reached intra and postoperatively with histopathological and culture findings. Herein, we report a young male patient that presented to our emergency department with signs and symptoms suggestive of intestinal obstruction. Intraoperatively, a thin membrane encasing the small bowels with transmesenteric herniation noted with a clinical diagnosis of SEP as the primary cause of obstruction. Postoperatively, a definitive cause was identified by histopathological findings and specimen culture. The report of this case, as one of the few male patients with this entity in the published literature, highlights the notion that surgeons should be vigilant to keep in mind this extremely rare cause while managing a patient with intestinal obstruction.

**Keywords:** Cocoon, Intestinal Obstruction, *Mycobacterium bovis*, Sclerosis, Tuberculosis.

Partial or complete encasement of variable length of the peritoneal organs predominantly the small bowel by a fibro-collagenous membrane resulting from chronic inflammation of the peritoneum either primary or secondary that gives the appearance of a cocoon is a rare cause of mechanical bowel obstruction. The primary (idiopathic) type of Sclerosing Encapsulating Peritonitis (SEP) described by Foo et al in adolescent girls, with no known etiology proposed to cause immunologic damage to the peritoneum due to retrograde menstruation and infection via fallopian tubes [1] while the secondary variant having known underlying cause, that include continuous peritoneal dialysis, abdominal tuberculosis, certain drugs like practalol, asbestos, methotrexate and various other uncommon etiologies like, cirrhosis, recurrent peritonitis, endometrioma, abdominal trauma, HIV, systemic lupus erythematosus, *Mycobacterium bovis* [2].

Herein, we report a case of intestinal obstruction with internal herniation in a young male patient with no definitive cause found preoperatively while postoperatively, definitive diagnosis reached as *Mycobacterium bovis* induced SEP. The treating surgeon should keep in mind this extremely rare cause while treating a patient with bowel obstruction and to reach an underlying predisposing factor to avoid morbidity and mortality.

### CASE REPORT

A 30-years-old otherwise healthy Egyptian male farmer presented to the emergency department (ED) of our secondary care facility with a history of colicky abdominal pain which was intermittent in nature with recurrent vomiting for the last two weeks. The patient travelled to Saudia a week before from his native country where he was treated for Bilharziasis as he visited the hospital for the same complaint. The doctor treated him for Bilharziasis empirically, that may be due to the common occurrence of Bilharziasis in that community. The patient visited our ED as his condition worsened with severe central abdominal pain, intermittent bilious vomiting, abdominal distension, and constipation.

On examination; a young ill-looking mildly dehydrated male patient with vitals as; pulse-84/min, respiratory rate 19/min Temp-37.8C<sup>0</sup> and blood pressure 130/70mmHg. Abdominal examination shows a moderately distended abdomen with generalized tenderness more marked at the central and lower abdomen. A provisional diagnosis of intestinal obstruction was made.

Laboratory workup shows; Hemoglobin-13.20g/dL, leucocyte-count-9770/mm<sup>3</sup>, platelets-191,300/mm<sup>3</sup>, alkaline phosphatase-51u/L, total bilirubin-14.90umol/L, direct bilirubin-2.30umol/L, creatinine-73umol/L, random blood sugar level-5.8mmol/L, serum Na<sup>+</sup>-13.40mmol/L, K<sup>+</sup>-3.40mmol/L. Plain

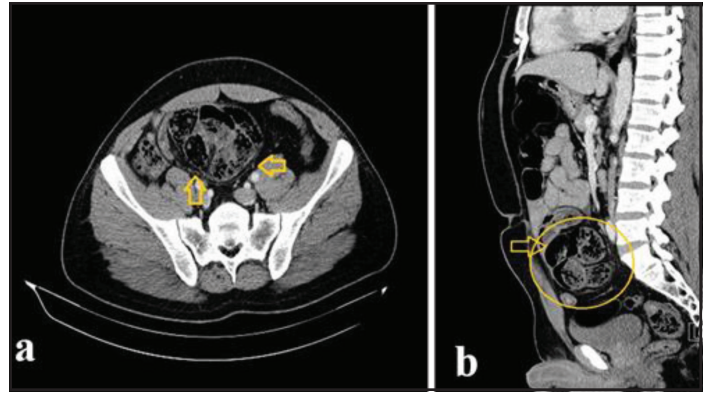


**Figure 1: Plain abdominal radiograph with small bowel dilatation and multiple air-fluid levels.**

abdominal radiograph shows multiple air-fluid levels with small bowel dilatation suggestive of intestinal obstruction (Fig. 1). Contrast-enhanced computed tomography (CECT) scan axial view (Fig. 2a) of the lower abdomen shows multiple gas-containing small bowel loops encapsulated within a sac-like structure (arrowheads) giving a cauliflower-like appearance, seen medial to the caecum and associated dilated proximal bowel with air-fluid levels due to intestinal obstruction. Sagittal view (Fig. 2b) of the abdomen shows a cluster of ileal loops encapsulated in a thin membrane-like sac (arrow) that occupies lower abdomen, likely internal hernia through ileocolic or other aperture.

In ED, the patient kept nil by mouth, a nasogastric tube inserted and IV fluid resuscitation was done. The patient prepared for emergency exploratory laparotomy based on clinical and radiological findings. Intraoperative findings were; partial encapsulation of whole small bowel by a fibro-collagenous thin membrane (Fig. 3a) with herniated dilated loops, thus a clinical diagnosis of sclerosing encapsulating peritonitis was made. Adhesions (Fig. 3b) relieved by finger sweeping and loops separated without resection as there were no signs of vascular compromise or gangrene. Omental and peritoneal specimen were obtained for the histopathological and culture evaluation.

The patient did well during a postoperative stay in the surgical unit and discharged on 8<sup>th</sup> postoperative day with a week later follow-up in surgical OPD. The patient was doing well with respect to surgery, in the follow-up period. The histopathology report of the taken specimen shows a caseating epithelioid cell granuloma and the organism identified as *M. bovis* by tissue culture. Postoperatively, a detailed questioning revealed the consumption of unpasteurized milk from their cattle in his farm. On the basis of clinical coupled with histopathological and culture confirmation, a definitive diagnosis of *Mycobacterium bovis* induced SEP was made. Upon confirmation of the definitive cause postoperatively, the patient was put on anti-tuberculous regime by the local TB hospital and followed on regular intervals with good improvement.



**Figure 2: Contrast-enhanced CT scan (a) axial view; (b) sagittal view of the lower abdomen.**

## DISCUSSION

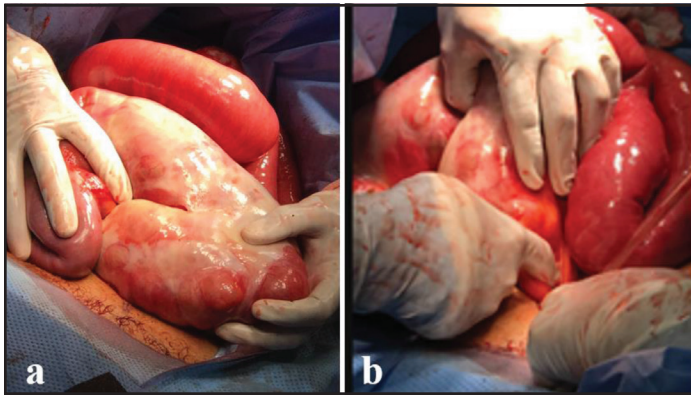
The primary (idiopathic) type of SEP was coined by Foo et al [1] in 1978 and termed as abdominal cocoon syndrome. The secondary form which is more common was first reported and termed ‘peritonitis chronica fibrosa incapsulata’ by Owtschinnikow [3] in 1907. The condition later termed by Deeb et al. [4] as SEP and encapsulating peritoneal sclerosis (ESP) [5].

Bowel obstruction is a commonly seen entity in surgical practice with various etiologies. Preoperatively, finding the cause is difficult and usually identified intra or postoperatively following laparotomy and biopsy findings. SEP, a chronic inflammatory condition in which the peritoneal organs, more commonly the small intestines are encased in a thick fibrous sheet is divided into primary (idiopathic) or secondary based on the underlying cause [6,7,8-11].

Another condition called peritoneal encapsulation (PE), a developmental anomaly characterized by an accessory thin mesothelial membrane encasing partially or in the whole small bowel, derived from the yok sac peritoneum in early fetal life. The condition first described by Cleland in 1868, is asymptomatic predominantly and detected during laparotomy for some other reason [12].

Extrapulmonary tuberculosis (EPTB) accounts for 15-20% cases with 3.0-6.7% of abdominal and 1.0 - 6.1% of peritoneal involvement of all cases EPTB [13-16]. Various forms of abdominal TB include luminal, visceral, lymph nodal and peritoneal based on the pattern of involvement [16]. Commonly involved sites of gastrointestinal tuberculosis include ileocecal junction, jejunum, and colon with esophagus, stomach, and duodenum are the least affected parts. Peritoneal TB involvement usually described as, dry adhesive type, wet ascitic type and fibrotic fixed type with loculated ascites and omental involvement [13]. In the available literature, few cases of abdominal cocoon due to tuberculosis has been reported [17] while McLeod et al. recently reported a case of *Mycobacterium* induced SEP with intra-abdominal fistulas in a 69-years-old Canadian man that was initially missed by extensive evaluation [18].

A rare fourth form with an overlap of the aforementioned three forms lead to adhesion and fibro-collagenous sheet formation like a cocoon encasing all or part of the bowel that may present with



**Figure 3: (a) Intraoperative findings of fibro-collagenous layer over the intestinal loops; (b) Fibro-collagenous layer separation by finger sweeping.**

bowel obstruction and abdominal mass [16,19]. Clinical features of abdominal TB depend on site of involvement with the most common early presenting features include abdominal pain, fever, weight loss, ascites and acute or sub-acute intestinal obstruction due to fibro-collagenous sheet formation as a late feature of the disease and the patient presents with signs and symptoms of intestinal obstruction as seen in our case [20-21].

Plain X-ray abdomen findings are non-specific and indicate features of intestinal obstruction such as dilated bowel loops with air-fluid levels [22]. Computed tomographic (CT) is the primary imaging modality with findings of peritoneal thickening, mural thickening, loops fixation, signs of obstruction, tethering, ascites, and loculated fluid collections [23] while small bowel obstruction, central position, clustering with mass effect on adjacent organs and engorged stretched mesenteric vessels all suggestive of internal herniation [24]. In a review article of Rastogi [25], the author concluded that abdominal cocoon secondary to abdominal tuberculosis can be diagnosed with confidence using various radiological investigations. Meng *et al* [26] in a recent study concluded that characteristic CT findings coupled with various biochemical parameters play a vital role in tuberculous abdominal cocoon diagnosis. The differential diagnosis includes congenital peritoneal encapsulation, internal herniation, peritonitis carcinomatosa, pseudomyxoma peritoneal mesothelioma and other causes of bowel obstruction [27-28].

Treatment of choice is relieving the obstruction and release of adhesions through exploratory laparotomy [29] or laparoscopic excision [30-31] that is rarely a preferred approach. The underlying cause needs to be sorted out to plan case-specific management.

## CONCLUSION

*Mycobacterium bovis* induced SEP leading to mechanical bowel obstruction with or without complications is rarely encountered in clinical practice. Definitive diagnosis is challenging in the pre-operative period and usually missed. A treating surgeon must reach a correct diagnosis with definitive

cause, as to plan case-specific management for the patient to avoid the undesired outcome.

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