

## Case Report

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# Case report of sclerosing encapsulating peritonitis secondary to tuberculosis

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## ABSTRACT

Sclerosing encapsulating peritonitis (SEP) is a visceral encapsulation syndrome of inflammatory origin, seen as an infrequent cause of intestinal obstruction in young patients. It is a condition characterised by complete encapsulation of small bowel loops by a fibrocollagenous membrane, leading to cocoon formation. Histologically, the membrane is composed mainly of organised fibrin, probably derived from the plasma exudation of peritoneal microvasculature. Clinical presentation is related to the development of altered gut motility, resulting in abdominal pain and features of intestinal obstruction. We here discussed about a young patient who presented with features of intestinal obstruction and diagnosed with SEP, secondary to abdominal tuberculosis and was then treated surgically. SEP as a cause of intestinal obstruction is a rare life threatening entity encountered in day to day practice. Definitive diagnosis of this condition is challenging in the pre-operative period and is usually missed and a high index of suspicion is required.

**Keywords:** Encapsulating peritonitis, Tuberculosis, Cocoon

## INTRODUCTION

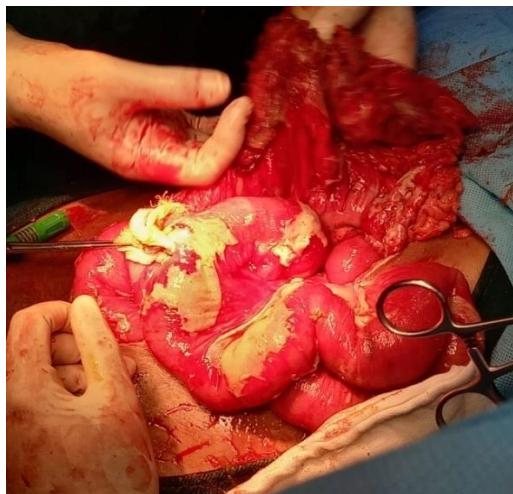
SEP is a rare disease entity, characterised by total or partial encasement of the small bowel loops by a thick fibrous sac, resulting in bowel obstruction. It was first described in 1907 by Owtchinnikow as peritonitis chronic fibrosa incapsulata.<sup>1</sup> This condition was later termed as SEP by Deeb et al.<sup>2</sup> In 1921, Winnen reported the first case of SEP and termed it zuckergussdarm, translating as icing gut, due to the presence of whitish membrane covering the intestinal loops.<sup>3</sup>

This was believed to be due to chronic intra-abdominal fibro inflammatory process, that resulted in formation of fibrous tissue sheets that covered and fixed the bowel loops, thereby compromising its motility. This eventually resulted in formation of marbled, leathery, thick fibro collagenous sheet like structure that enveloped the small intestines in form of cocoon. The aetiology remains unknown and is considered to be multifactorial.

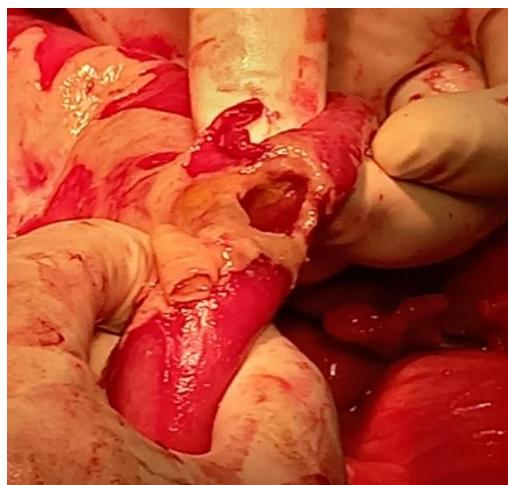
Diagnosis is delayed owing to the rarity of the disease and non-specific symptoms and laboratory findings.

## CASE REPORT

A 27 year old male brought to the ED of our institute with complaints of abdominal pain, abdomen distension and obstipation for one day. He was recently diagnosed with miliary tuberculosis and started on ATT. Otherwise, he had no significant past medical history or any comorbid illness. On examination, an afebrile, thin built and malnourished young male, in dehydrated state with vitals PR 103 /min, BP 110/70 mmHg, RR 22 /min, SpO<sub>2</sub> 98% in RA, CBG 140 mg/dl, abdominal girth 75 cm was seen. Abdominal examination showed tense, distended abdomen with diffuse tenderness throughout the abdomen with absent bowel sounds. Digital rectal examination showed fecal pellets. Other system examination were normal. A working diagnosis of acute intestinal obstruction was made.



**Figure 1: Dense membrane and flakes over the surface of small bowel loops.**



**Figure 2: Perforation of size 2x1 cm over antimesenteric border of ileum.**

Initial laboratory workup showed anemia with leukocytosis and hypoalbuminemia. Chest X-ray showed diffuse heterogenous opacities. Contrast enhanced CT (CECT) abdomen showed opacification of bowel loops, loculated peritoneal collections in perirectal and mesenteric spaces with bowel wall thickening and multiple sub centimetric lymph nodes in the mesenteric space.

The patient was resuscitated with intravenous fluids, blood transfusion and prepared for emergency laparotomy based on clinical and radiological findings. After explaining the risks involved and obtaining written consent, abdomen opened through midline incision and intra-operative findings were: adhesions between small bowel loops, forming conglomerate mass, resembling abdominal cocoon; tiny flakes distributed throughout the surface of small bowel loops; purulent exudate of about 50 ml admixed with about 100 ml ascitic fluid;

perforation of size 2x1 cm, about 80 cm from ileocecal junction over the antimesenteric border of ileum, concealed by the dense adhesions (Figure 2).

The adhesions were released by finger sweeping and loops separated, thorough wash given, unhealthy omentum was clamped and cut, the perforation identified on removal of adhesions, segment of ileum along with perforation resected and managed with double barrel ileostomy. Resected specimen was sent for histopathology examination and ascitic fluid was sent for analysis. Post-operatively, patient was re-started on ATT and managed with IV antibiotics and total parenteral nutrition. On post operative day 3, patient developed sudden respiratory distress and was intubated. He developed clinical features of TB meningitis and succumbed to it on post-operative day 10.



**Figure 3: Resected unhealthy omentum covered with whitish membrane and flakes.**

Histopathology examination showed epithelioid granulomata composed of epithelioid cells, lymphocytes, histiocytes and Langerhans type of giant cells with areas of necro inflammation and congested blood vessels, typical of granulomatous lesion, possibly of tuberculous etiology. Ascitic fluid analysis showed elevated adenosine de-aminase enzyme with weakly detected *Mycobacterium* tuberculosis organism.

## DISCUSSION

SEP is a rare cause of inflammation of the peritoneum and intestinal obstruction, resulting from encasement of variable amounts of small bowel by a fibro-collagenous membrane.<sup>4</sup> SEP can be primary (idiopathic) or secondary to an identified underlying cause. The primary (idiopathic) variant of SEP was termed by Foo et al in 1978 as abdominal cocoon syndrome.<sup>5</sup> This was found to be common among adolescent girls, with no known cause, proposed to be caused by cell mediated immunologic damage to the peritoneum due to retrograde

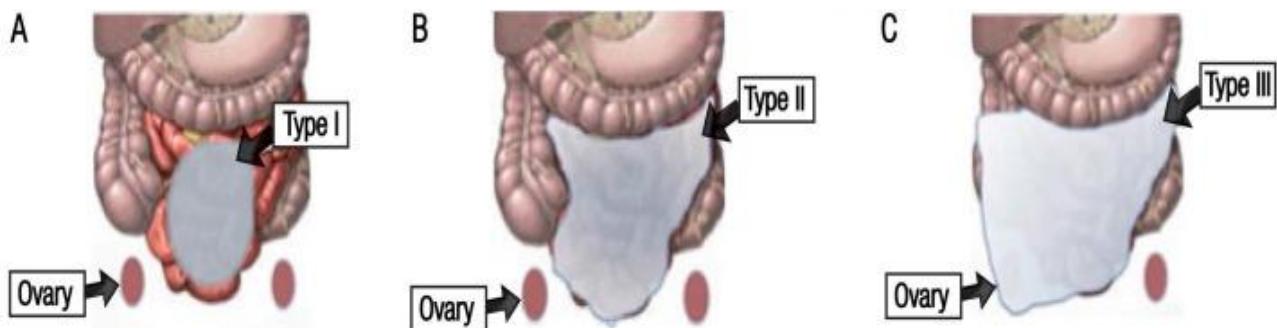
menstruation and infection through the fallopian tubes. However, since this condition had also been reported in males, premenopausal females and children, there was little support to this hypothesis.

Abdominal cocoon syndrome was divided into three types based on extent of membrane encasement. In type I and II, the small intestine loops were partially or completely covered by membrane, respectively and in type III, the membrane enclosed other structures like appendix, ovaries, cecum, ascending colon, stomach and liver apart from small intestine (Figure 4).<sup>6</sup>

The classification system proposed by Nakamoto et al depending on clinical features in patients with peritoneal dialysis associated SEP, divided it into 4 phases, namely pre sclerosing encapsulating peritonitis; inflammatory phase; encapsulation; and chronic inflammatory stage.<sup>7</sup>

**Table 1: Phases of SEP.**

Phases		
<b>Phase 1</b>	Pre sclerosing encapsulating peritonitis	Asymptomatic with mild ascites and no inflammation
<b>Phase 2</b>	Inflammatory stage	Symptomatic with nausea and diarrhoea consistent with partial encapsulation of bowel; mild inflammation with fibrin exudation seen
<b>Phase 3</b>	Encapsulation	Associated with mild to severe inflammation; symptomatic of bowel obstruction due to complete encapsulation of bowel loops by membrane
<b>Phase 4</b>	Chronic inflammatory stage	Patients have complete bowel obstruction due to thickening of the encapsulating fibro-collagenous cocoon



**Figure 4: Types of abdominal cocoon syndrome.**

The secondary causes included long standing peritoneal dialysis, abdominal tuberculosis, certain drugs like propranolol (beta blocker), methotrexate, asbestos, cirrhosis, recurrent peritonitis, abdominal trauma, HIV, endometrioma, systemic lupus erythematosus, familial Mediterranean fever, sarcoidosis, gastrointestinal malignancy, fibrogenic foreign body, protein S deficiency, liver transplantation, luteinized ovarian thecoma, the use of povidone iodine for abdominal washout.<sup>8</sup> The above cited causes induced peritoneal inflammation, leading to fibrogenesis and formation of the dense membrane, characteristic of SEP.

One of the cited secondary causes of SEP included abdominal tuberculosis. Extra-pulmonary tuberculosis accounted for 15-20% cases, with 3.0-6.7% abdominal and 1.0-6.1% peritoneal involvement. Commonly involved sites in gastrointestinal tuberculosis were ileocecal junction, jejunum and colon with esophagus, stomach and duodenum being least affected. Various forms of abdominal TB included luminal, visceral, lymph nodal and peritoneal based on pattern of involvement.<sup>9</sup> Peritoneal TB was described as dry adhesive type, wet ascitic type and fibrotic fixed type with loculated ascites and omental involvement.<sup>10</sup> Clinically, the incidence of

acute small bowel obstruction was higher in patients with tubercular cause of SEP. Conservative management generally failed, due to the higher incidence of inter-bowel adhesions and fibrosis in tubercular form, in addition to the fibro-collagenous membrane.

Clinical diagnosis of SEP was facilitated by presenting symptoms of the patient, pre-existing risk factors and importantly, high index of suspicion. Radiological studies used to diagnose SEP included abdominal X-ray, barium study, ultrasound, CECT and if required, MRI. Abdominal X-rays were nonspecific demonstrating multiple air-fluid levels and dilated bowel loops, suggestive of intestinal obstruction. Ultrasound abdomen showed trilaminar membrane, characteristic of SEP.

Barium study showed central clumping of the gut, described as cauliflower pattern or Accordion sign, due to the membrane encasement. CECT was the most reliable and sensitive method to diagnose SEP. Findings included peritoneal thickening (100%), calcifications (70%), peritoneal enhancement (50%), congregated small bowel loops in the centre of abdomen (60%) and loculated fluid collections (90%).<sup>11</sup> However, a definite diagnosis was made intra-operatively only.

Treatment of SEP included elimination of the fibrogenic agent and treatment of the underlying cause. Bowel rest and total parenteral nutrition have shown to be effective. Medical treatment regimens based on corticosteroids have been recommended based on its effectiveness against fibrotic diseases.<sup>12</sup> Surgical exploration with adhesiolysis and dissection of the membrane remained the hallmark of treatment of SEP. Options included membrane excision with adhesiolysis or for patients with gut injury, resection and anastomosis with or without a protective enterostomy was done.<sup>13</sup>

An integral part of the treatment was complete excision of the membrane, which ensured reduction in recurrence rate. Fibrotic membranes which covered the coiled intestinal loops like thick plastic bag may pose technical challenge and separating these membranes from underlying bowel may need multiple longitudinal and transverse incisions.<sup>14</sup> These incisions may facilitate the stripping of the membrane to allow the underlying intestine loops to regain their length and function. But the potential for iatrogenic complications was high. Other intra-operative findings like enlarged and caseating lymph nodes, mesenteric abscess and tubercles over the bowel serosa may suggest a tubercular etiology.<sup>15</sup>

## CONCLUSION

SEP is a rare clinical entity and is often encountered unexpectedly in patients with acute intestinal obstruction. High index of clinical suspicion along with radiological investigations, particularly CT imaging of the abdomen, in susceptible patients helps in achieving a preoperative diagnosis. Surgery is vital in management of this condition. Careful dissection and removal of the membranous adhesions leads to recovery.

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