

# Case Report

## Peritoneal Tuberculosis Mimics Peritoneal Encapsulation Syndrome

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**Background:** Peritoneal encapsulation (PE) syndrome is a rare congenital condition with an accessory peritoneal sac covering the small bowel. PE is mostly reported to be asymptomatic and detected incidentally, however, it can present as a rare cause of gut obstruction if there is peritoneal inflammation as our case report.

**Case Report:** A Thai 47-year-old man presented with clinical of small bowel obstruction in virgin abdomen. Physical examination revealed abdominal distension with large palpable cystic lesion in abdomen. CT scan revealed small bowel loops within a thin membranous sac in the mid-abdomen containing fluid and a disproportion point at duodenojejunal junction. Exploratory laparotomy was performed, and found a peritoneal cyst in the mid-abdomen containing entire small bowel and clear yellow fluid. Stomach and the large bowel were located outside of the cyst. There was a band of the peritoneal membrane causing obstruction at the duodenojejunal junction. The cyst was opened, and its membrane and fluid were sent for pathological, cytological and microbiological examination. Obstruction point was released. Post-operative course was uneventful. The pathological report revealed chronic granulomatous inflammation; tissue polymerase chain reaction for tuberculosis was found positive. The patient was treated with anti-tuberculosis drugs and had no further symptom of gut obstruction.

**Conclusion:** Peritoneal tuberculosis is likely to be an aggravating factor for gut obstruction in peritoneal encapsulation syndrome, which is a congenital condition, by causing the peritoneal thickening and obstruction at duodenojejunal junction.

**Keywords:** Peritoneal encapsulation, Sclerosing encapsulating peritonitis, Cocoon abdomen, Peritoneal tuberculosis, Intestinal obstruction

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Peritoneal encapsulation (PE) is a rare congenital anomaly, presenting with an accessory peritoneal membrane covering the small bowel. This has been described interchangeably with sclerosing encapsulating peritonitis (SEP) and cocoon abdomen (CA). However, these three conditions are different in pathophysiology and natural course of diseases. PE is mostly asymptomatic and diagnosed incidentally during surgery or autopsy<sup>(1-4)</sup>. Clinical presentation with gut obstruction is rare. Herein, we present a case report of small intestinal obstruction, which is caused by peritoneal tuberculosis in peritoneal encapsulation syndrome.

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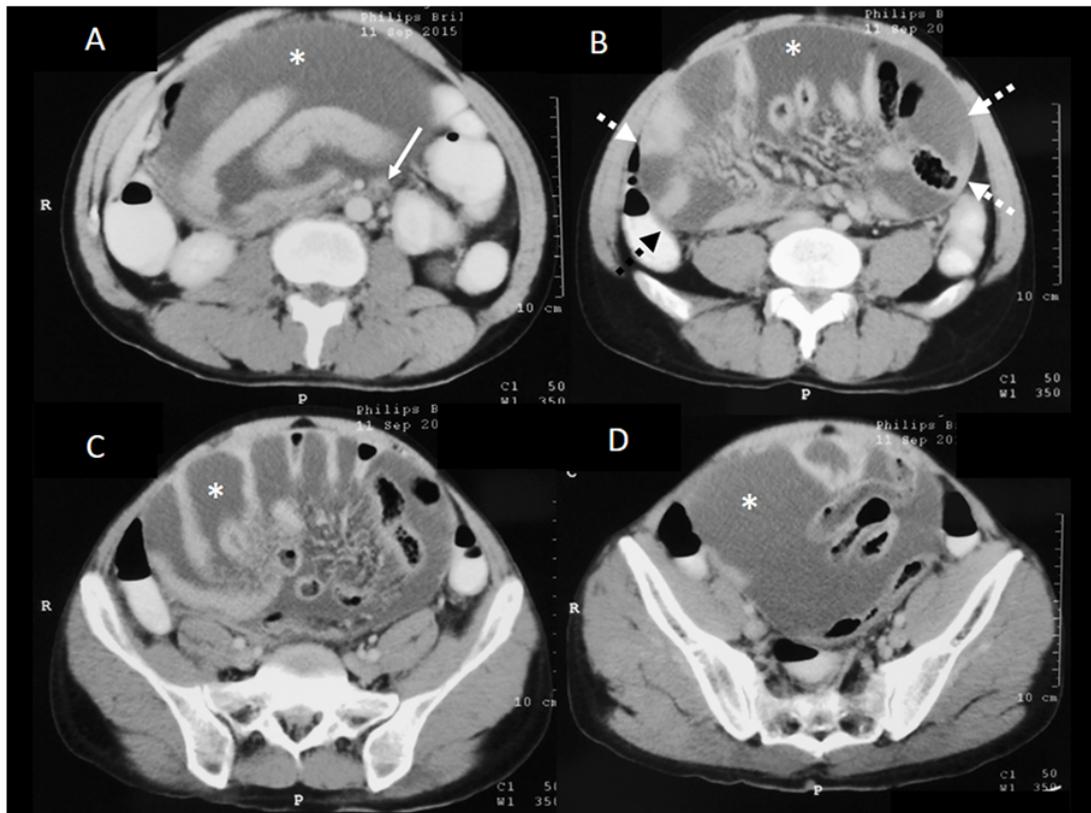
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### Case Report

A 47-year-old Thai male presented with severe abdominal pain with obstipation for three days. He had a history of intermittent colicky abdominal pain without constipation or obstipation for two weeks. He was previously healthy and had no history of previous abdominal surgery. The patient was admitted to a local hospital. Physical examination revealed abdominal distension with large palpable cystic lesion in mid-abdomen. Abdomen was otherwise soft, no tenderness and hyperactive bowel sound. Conservative treatments with nasogastric tube decompression, nothing per oral and intravenous fluid were given. Plain abdominal radiography showed generalized small bowel dilatation with different height of air-fluid levels in the same loop and collapsed large bowel. Small bowel obstruction was suspected. Laboratory investigations were all within normal limits. Abdominal CT scan revealed edematous small bowel loops within a surrounding thin membranous sac in the mid-abdomen containing fluid. The large bowel was collapsed and laid outside of the



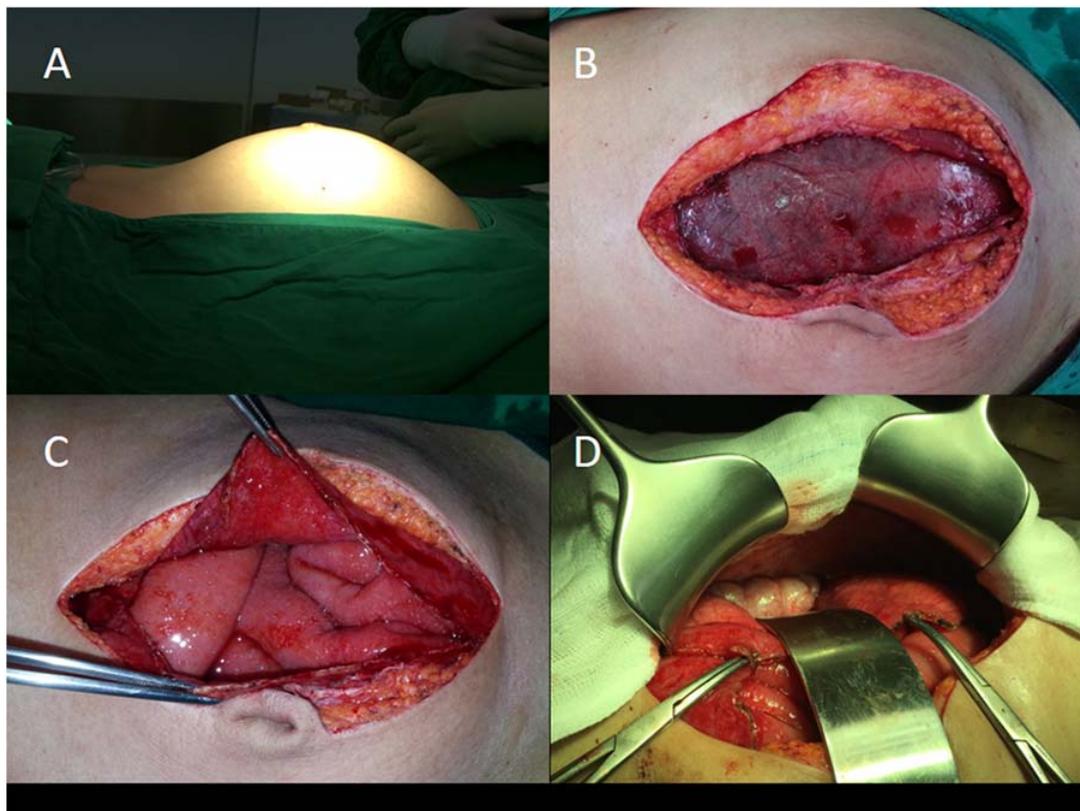
**Fig. 1** The abdominal CT scan of the patient revealed an encapsulated peritoneal sac (\*) in the central abdomen, containing serpentiniform pattern of small bowel loops, mesentery and fluid. The small bowel wall was edematous. Colon were outside the sac, and there was no ascites outside the sac. The dotted arrows show the wall of the encapsulated peritoneal sac. The white arrow shows the disproportion at the duodenojejunal junction where the bowel passes into the peritoneal sac.

sac. Duodenum was dilated with disproportion at duodenojejunal junction (Fig. 1).

The patient was referred to Siriraj Hospital for further investigation and treatment. From the clinical presentation and the CT scan, the provisional diagnosis of peritoneal encapsulation syndrome with complete small bowel obstruction was suspected. An exploratory laparotomy was performed. The laparotomy revealed a peritoneal cyst in the middle of abdomen, approximately 20x30 cm in size, extending from transverse mesocolic root superiorly, to pelvic cavity inferiorly, and between ascending and descending colon laterally. The cyst contained jejunum, ileum and approximately 1,000 mL of clear yellow serous fluid. There were multiple small nodules along small bowel serosa, parietal peritoneum and the cyst wall (Fig. 2). The duodenum was dilated with disproportion point at duodenojejunal junction. The small bowel appeared inflamed but viable, while the large bowel, found

outside of the cyst, appeared healthy. The cyst was opened, and the cyst fluid was sent for cytology and microbiological examination. The cyst wall was partially excised and sent for pathological and microbiological examination. The adhesion around duodenojejunal junction was lysed and the duodenojejunal junction was freely released. The content was confirmed to have passed through the obstruction point.

The postoperative course was uneventful. The patient could resume diet on postoperative day 2 and be discharged home on postoperative day 8. Gram and AFB stains from the cyst fluid and the cyst wall tissue were all negative. Also, aerobic bacterial culture from the cyst fluid and the cyst wall tissue were no growth. The pathological report of the cyst wall was peritoneal tissue with chronic granulomatous inflammation. Although acid-fast bacilli or fungi were not identified from AFB, PAS and GMS stains, mycobacterial infection could not be excluded. Therefore, the polymerase chain



**Fig. 2** Intraoperative findings of the patient. A) Shows abdominal distension, but only in the central abdomen where the cyst lied. B) Shows an accessory peritoneal sac in the middle of abdominal cavity after exploratory laparotomy. C) Shows the small bowel that lied inside the sac. The small bowel were edematous and inflamed with multiple small nodules on the serosa. D) Shows left-side colon which lied outside the sac and appeared healthy.

reaction (PCR) for tuberculosis (TB) was further requested, and found positive for *M. tuberculosis* complex.

After confirmation of disseminated TB to peritoneum, an infectious disease physician was consulted. A four-drug regimen of anti-tuberculosis medication, consisting of 300 mg/d of isoniazid, 600 mg/d of rifampicin, 1,250 mg/d of pyrazinamide and 800 mg/d of ethambutol, was started. The patient recovered well, and had no further symptom of abdominal pain or gut obstruction.

### Discussion

Peritoneal tuberculosis is one of the most common presentations of extra-pulmonary tuberculosis, especially in the endemic area of tuberculosis. The main clinical presentation is ascites<sup>(5)</sup>. However, it can sometimes be presented with gut obstruction, which is mostly from inflammation at distal ileum causing luminal narrowing and distal small bowel

obstruction. Proximal small bowel obstruction at duodenojejunal junction is rare.

In our patient, gut obstruction was aggravated by disseminated tuberculosis infection as confirmed by PCR. The patient has an accessory peritoneal sac since birth, but without any symptom. Gut obstruction made him seek medical attention and subsequently be diagnosed with PE. Peritoneal tuberculosis causes thickening of peritoneal membrane including an accessory peritoneal membrane that forms the sac. Dense thickening fibrous tissue causes obstruction at the entry of the small bowel in to the accessory sac at the duodenojejunal junction<sup>(2)</sup>. This requires urgent surgery with excision of the membrane, releasing a band obstructing the bowel or lysis of adhesions between bowel loops. Small bowel resection is usually unnecessary, unless there is an evidence of bowel ischemia.

Peritoneal encapsulation syndrome is a rare congenital condition with an accessory peritoneal cyst

wall covering the small bowel. It was first described by Cleland in 1868<sup>(6)</sup>. Until now, there are less than 20 cases reported in literature, mostly diagnosed incidentally<sup>(3)</sup>. The pathophysiology of PE comprises of abnormal return of midgut loop in to the abdominal cavity during the 12<sup>th</sup> week of development<sup>(2)</sup>. Thus, the small bowel is covered by the original dorsal mesentery, which normally forms the transverse mesocolon, presenting as an accessory peritoneal sac<sup>(7)</sup>. The boundaries of the peritoneal sac are the ascending and descending colon laterally, transverse colon superiorly, and parietal peritoneum inferiorly. The peritoneal membrane covers the entire small bowel, from duodenojejunal junction to ileocecal valve. Stomach and large bowel, in turn, stay outside of this accessory peritoneal sac<sup>(8)</sup>.

Patients with PE are usually asymptomatic, but may occasionally present with gut obstruction<sup>(2,8)</sup>, as in our patient. In physical examination, asymmetrical and fixed abdominal distension that does not vary with peristalsis may be noted in PE with gut obstruction. On palpation, abdomen will have a difference in consistency, where a dense fibrous capsule is firm and distended bowel is soft<sup>(2)</sup>. The pre-operative CT scan may show a characteristic serpentiniform pattern of small bowel in a fluid-fill sac in the middle of abdomen. Additional findings are U-shaped small bowel loops, slowed transit, intestinal wall thickening and signs of obstruction<sup>(8)</sup>. At exploratory laparotomy, the characteristic of PE is an accessory peritoneal sac encasing the entire small bowel. Reactive fluid is usually found in the sac. Stomach, duodenum and large bowel are all outside the accessory peritoneal sac.

There are two additional conditions of peritoneum frequently confused and sometimes used interchangeably and falsely: sclerosing encapsulating peritonitis (SEP) and cocoon abdomen (CA). PE is a congenital condition, whereas SEP and CA are acquired diseases<sup>(2)</sup>. Moreover, PE has good prognosis with low rate of recurrence, while SEP is associated with 40 to 80% mortality from intestinal necrosis, anastomotic leakage and enterocutaneous fistula<sup>(9)</sup>.

SEP was first described by Owtschinnikow in 1907<sup>(4)</sup>. This is a complication of chronic ambulatory peritoneal dialysis. After prolonged peritoneal dialysis, the peritoneum will be chronically inflamed by acetate in the dialysate and antiseptic used during bag changes. It is characterized by a thick greyish-white fibrous membrane covering all intraperitoneal viscera. Beta-blocker, recurrent peritonitis, ventriculo-peritoneal shunt, peritoneovenous shunt, sarcoidosis and

systemic lupus erythematosus are also believed to be causative factors<sup>(2)</sup>. Besides surgery, conservative treatment has been reported with success using corticosteroids, immunosuppressive agents and tamoxifen.

CA is another entity that resembles PE. It was first described in 1978 by Foo et al<sup>(10)</sup>. This was believed to be associated with retrograde menstruation, however, there have been reported in both children and men. Therefore, the exact etiology of CA remained unknown. There is a case series reporting five cases of CA developing after orthotopic liver transplantation<sup>(11)</sup>. GI follow-through study would show typical characteristic of serpentine configuration of dilated small bowel within a cocoon-like structure. Intra-operatively, the small bowel would be found totally or partially coiled up in serpentine-like fashion, and encased in a thick dense white membrane.

## Conclusion

PE is a rare congenital condition characterized by an accessory peritoneal sac covering the small bowel. Even though PE is mostly asymptomatic, gut obstruction can occur if there is a peritoneal disease such as TB peritonitis. We present a case of gut obstruction from a rare cause of PE complicating with TB peritonitis.

## What is already known on this topic?

Peritoneal tuberculosis can present with ascites and sometimes gut obstruction. Peritoneal encapsulation syndrome is a congenital abnormality with an accessory peritoneal sac covering the small bowel, and is normally asymptomatic.

## What this study adds?

Gut obstruction can occur at duodenojejunal junction in peritoneal encapsulation syndrome when there is an inflammation process of the peritoneum such as peritoneal tuberculosis.

## Potential conflicts of interest

None.

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## วัฒนธรรมของท้องที่แสดงอาการเหมือนถุงเยื่อช่องท้องหุ้มลำไส้เล็ก

สุทธิวิชัย โควิกภัย, ประวิทย์ โมลิตะมงคล, ยงยุทธ สิริวัฒนอักษร

**ภูมิหลัง:** Peritoneal encapsulation เป็นความผิดปกติแต่กำเนิดชนิดหนึ่งที่พบได้ไม่บ่อย โดยพบมีถุงเยื่อช่องท้อง คลุมลำไส้เล็ก โดยทั่วไปมักจะพบโดยบังเอิญแต่ก็อาจพบเป็นสาเหตุของการเกิดลำไส้อุดตันได้ดังเช่นในรายงานฉบับนี้

**รายงานผู้ป่วย:** ผู้ป่วยชายไทยอายุ 47 ปี มีอาการของลำไส้เล็กอุดตันโดยผู้ป่วยไม่เคยได้รับการผ่าตัดช่องท้องมาก่อน การตรวจร่างกายพบท้องอืดร่วมกับคลื่นพุงน้ำขนาดใหญ่ที่บริเวณกลางท้อง การตรวจเอกซเรย์คอมพิวเตอร์ช่องท้องพบลำไส้เล็กขดอยู่ในถุงน้ำขนาดใหญ่ ผู้ป่วยได้รับการรักษาโดยการผ่าตัดซึ่งพบว่ามีถุงน้ำขนาดใหญ่ในช่องท้องหุ้มรอบลำไส้เล็กทั้งหมด กระเพาะอาหารและลำไส้ใหญ่อยู่นอกถุงน้ำนี้ ลำไส้อุดตันเกิดจากถุงน้ำนี้รั่วบริเวณ duodenojejunal junction ผู้ป่วยได้รับการผ่าตัดรักษาโดยตัดเปิดถุงน้ำนี้และตัดแผ่นเยื่อช่องท้องที่รัดลำไส้และได้นำแผ่นเยื่อของถุงน้ำและน้ำในถุงน้ำไปส่งตรวจทางพยาธิวิทยา เซลล์วิทยา และเพาะเชื้อตรวจ หลังผ่าตัดผู้ป่วยมีอาการดีขึ้น ผลการตรวจทางพยาธิวิทยาพบว่าเยื่อของผนังถุงน้ำมีการอักเสบเรื้อรังชนิด chronic granulomatous และได้นำชิ้นเนื้อไปตรวจ polymerase chain reaction พบว่ามีเชื้อไวรัสก่อโรคผู้ป่วยจึงได้รับการรักษาไวรัสและไม่มีอาการเรื่องลำไส้อุดตันอีกเลย

**สรุป:** Peritoneal encapsulation เป็นความผิดปกติแต่กำเนิดของเยื่อช่องท้องซึ่งส่วนใหญ่ไม่ก่อให้เกิดอาการ การติดเชื้อไวรัสก่อโรคจะเป็นสาเหตุที่ทำให้ผนังเยื่อช่องท้องหนาตัวขึ้นและเกิดภาวะลำไส้อุดตันในผู้ป่วยรายนี้