

A Case Report of Abdominal Cocoon Syndrome and Small Intestinal Bezoar

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ABSTRACT

Sclerosing encapsulated peritonitis (SEP), referred to as abdominal cocoon syndrome (ACS), is an uncommon clinical etiology of acute mechanical intestinal obstruction. It is characterized by the small intestine being partially or completely covered by a white fibrocollagen membrane layer that resembles a cocoon. Bezoars are solid formations primarily made up of undigested food remnants and are an uncommon cause of acute intestinal obstruction.

Case presentation: A 58-year-old female patient with no history of previous abdominal surgery and no comorbidities was admitted to our hospital with clinical symptoms of intestinal obstruction. Following the necessary imaging and examinations, an emergency diagnostic laparotomy was performed with the diagnosis of acute mechanical intestinal obstruction. Intraoperatively, ACS and a 3 cm sized bezoar was detected in the jejunum and completely obstructing the lumen. It was thought that this patient's ACS may have contributed to his chronic constipation and ultimately to the development of bezoar, thus causing acute mechanical intestinal obstruction.

Conclusion: Careful evaluation of the underlying causes is essential in patients with acute mechanical intestinal obstruction, particularly when clinical symptoms are non-specific and common risk factors are absent. Early identification of bezoars and timely intervention may reduce the need for surgery in selected cases. Especially in SEP cases, the surgical approach plays a key role in both diagnosis and treatment. Clinicians should be aware that although SEP and intestinal bezoars are individually rare, they can occasionally occur together, making intraoperative assessment and management crucial for a successful outcome.

Keywords: abdominal cocoon syndrome, bezoar, ileus, acute intestinal obstruction, emergency surgery

Introduction

Sclerosing encapsulating peritonitis (SEP) is an uncommon condition and marked by the presence of a thick, fibrous membrane that wraps around the small intestine and in some cases, also covers other abdominal organs such as the liver, stomach, or colon (1). This membrane forms a cocoon-like structure, which limits bowel movement and may cause blockage (2). There are two types of SEP. The first type is called abdominal cocoon syndrome (ACS), also known as primary or idiopathic SEP, where no clear underlying cause is identified. The second type is secondary SEP, which develops due to known conditions or factors. Patients with ACS may show different clinical symptoms. These can range from mild, recurring episodes of partial intestinal obstruction to sudden and severe obstruction. Although imaging methods such as contrast-enhanced CT

scans can help raise suspicion of SEP, the definitive diagnosis is usually confirmed during surgical exploration, where the characteristic fibrous membrane can be clearly observed (3, 4).

A bezoar is a mass formed from undigested material that accumulates in the gastrointestinal tract, most commonly in the stomach. Bezoars are typically associated with increased intake of indigestible substances, previous gastric surgery or impaired gastrointestinal motility (5). The prevalence of bezoar development is 0.4%, although the exact incidence varies depending on the type of bezoar (6). They develop when materials that cannot be digested, such as plant fibers, hair, medications, or milk curds, gather and grow over time. Bezoars are classified into four main types based on their composition: phytobezoars (plant material), trichobezoars (hair), pharmacobezoars (medications or capsules), and lactobezoars (milk products). Among these,

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phytobezoars are the most common and are usually made of indigestible parts of fruits, vegetables, seeds, or shells (7). Most patients with bezoars are asymptomatic; however, some may present with symptoms such as abdominal pain, early satiety, nausea, or vomiting. In rare cases, bezoars may lead to serious complications like gastrointestinal obstruction or perforation (5).

Case Report

A 58-year-old female patient with no history of abdominal surgery or known comorbidities presented to our hospital with complaints of abdominal pain, bloating, nausea, and vomiting that had persisted for one week.

On physical examination, her abdomen was distended, but there were no signs of peritonitis.

Laboratory findings were within normal limits.

A plain abdominal X-ray showed multiple air-fluid levels suggestive of small bowel obstruction (Figure 1). Further evaluation with contrast-enhanced abdominal computed tomography (CT) revealed a suspected bezoar causing complete obstruction in the small intestine. The oral contrast could not pass beyond the lesion, and marked dilation of the proximal small bowel loops was observed.

Based on these findings, the patient was taken to the operating room for emergency exploratory laparotomy with a preoperative diagnosis of acute mechanical intestinal obstruction. Intraoperatively, a transparent, membranous structure resembling the peritoneum was discovered. This structure was encapsulating the entire small intestine and partially covering the colon (Figure 2). The membrane was carefully excised and sent for histopathological analysis. Additionally, a firm, intraluminal mass measuring approximately 3 cm was identified 160 cm distal to the ligament of Treitz, completely obstructing the jejunal lumen (Figure 3). The bezoar was removed via an enterotomy performed on the jejunal loop, and the enterotomy site was subsequently closed in a standard fashion. The procedure was completed without any intraoperative complications.

The patient's postoperative course was uneventful. She tolerated oral intake on the second postoperative day and was discharged fully recovered on the fourth day after surgery.

The histological analysis of the membrane structure revealed a diagnosis of SEP.

Discussion

ACS, or idiopathic SEP, is even less common, particularly in patients without any history of abdominal surgery, peritoneal dialysis, or other known risk factors (8). In this case, the patient had no previous surgeries or comorbidities, which supports the diagnosis of primary SEP (9). The clinical manifestations of SEP encompass symptoms including episodes of intestinal blockage, abdominal discomfort, nausea, vomiting and weight loss (10). There are instances in which SEP can also manifest as acute abdominal syndrome (9). As is the case with our patient, the laboratory results are not specific.

Timely intervention, which can considerably affect prognosis and lower the risk of severe consequences, is made possible by early radiologic identification of these indications. Therefore, through increased diagnostic accuracy and precision, radiology plays a critical role in directing therapeutic decision-making and enhancing outcomes for patients with SEP. CT findings of SEP involve peritoneal thickness, contrast enhancement and peritoneal calcifications, ascites and the appearance of clustered small bowel loops or the intestine with a membrane structure can be identified (11). Our CT findings were negligible. Primarily associated with jejunal bezoar. CT imaging reveal no abnormalities indicative with SEP.

The diagnosis of SEP is still very challenging to make before surgery. In most cases, the diagnosis of SEP is only provided during exploratory surgery (2). Additionally, the diagnosis of SEP is usually an unexpected finding during intraoperative exploration because it is caused by evidence of intestinal blockage (9, 12). Small bowel loop entrapment and gathering are the results of peritoneal thickening and fibrosis. Adhesions may occur, and the small bowel is then positioned in the midline, this finding is called the "cocoon sign" or "cauliflower sign" (9, 13, 14). Surgical intervention is the definitive standard for the therapy of primary SEP (8, 15). Our intraoperative and histopathological results were consistent, the diagnosis of primary SEP and jejunal bezoar was confirmed.

Bezoars are uncommon causes of small bowel obstruction and are most often seen in patients with risk factors such as altered gastrointestinal motility, previous abdominal surgeries, excessive intake of high-fiber foods and insufficient mastication (16). However, none of these reasons were present in our patient's history. The main

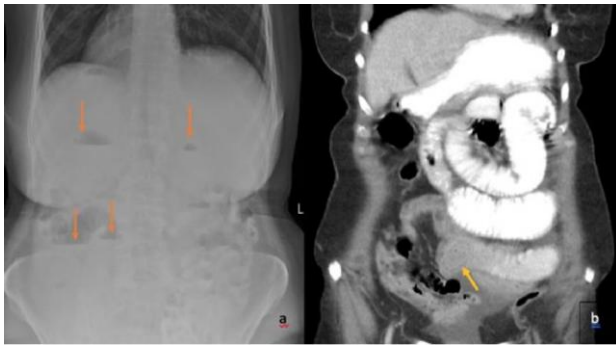


Fig. 1. The patient's preoperative abdominal X-ray (a) and CT scan (b) coronal plane

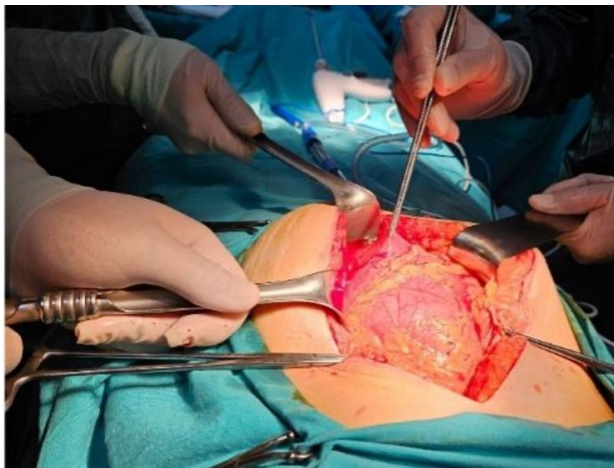


Fig. 2. The bright transparent membrane structure surrounding the small intestine

symptoms associated with bezoars include dyspepsia, abdominal pain. Although they are more commonly seen in the stomach, primary bezoars occurring in the small intestine can also cause symptoms and obstruction such as abdominal pain, nausea, and vomiting (16, 17). Our patient also had these symptoms.

Imaging tests, particularly CT are crucial in the diagnosis of bezoar (18) (19). Abdominal CT can identify the degree of obstruction and pinpoint the location of the bezoar.

The primary treatment for bezoar is conservative care; however, but postponing surgery may raise the risk of complications and death (17). Endoscopic removal of bezoars may be feasible in some cases; however, the majority require surgical intervention (19). Surgery was the only treatment option because our patient had signs of obstruction and the bezoar caused obstruction in the small intestines.

Interestingly, in this case the obstruction was not only caused by the SEP but also by a bezoar found in the jejunum. The coexistence of SEP with an intestinal bezoar is exceedingly rare and

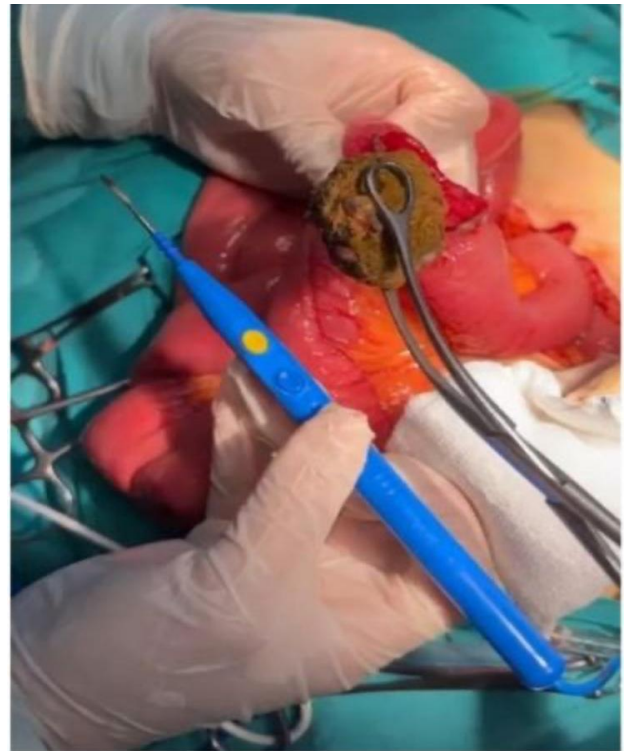


Fig. 3. Bezoar removed from 160 cm from Treitz

this may complicate the clinical picture and make preoperative diagnosis more challenging. SEP may have contributed to altered intestinal motility, creating a favorable environment for bezoar formation. The bezoar, in turn, exacerbated the intestinal obstruction, complicating the clinical scenario. Although abdominal CT helped identify the obstructing bezoar in this case, the SEP could only be confirmed during surgery (2). The surgical approach in this case was pivotal. The identification and excision of the fibrous membrane, along with the removal of the bezoar, were essential in resolving the obstruction and restoring normal intestinal function. This underscores the importance of considering SEP in the differential diagnosis of unexplained intestinal obstructions and the need for a high index of suspicion when managing such cases (20). The patient responded well to surgical treatment, including excision of the fibrous membrane and removal of the bezoar and had an uneventful recovery.

The coexistence of SEP and an intestinal bezoar is extremely rare and their nonspecific symptoms make preoperative diagnosis difficult. Therefore, in patients presenting with signs of bowel obstruction—especially in the absence of typical risk factors—clinicians should include both SEP and bezoar in the differential diagnosis.

Early surgical exploration remains essential for definitive diagnosis and effective treatment, helping to prevent severe complications and ensure a favorable outcome.

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Ethics Approval: Not applicable

Consent for Publication: Informed consent was obtained from this patient in the article.

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