CASE REPORT

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Abdominal Cocoon: A Rare Cause of Intestinal Obstruction. A Case Report

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Abstract

Abdominal cocoon, also known as sclerosing encapsulating peritonitis (SEP) is a relatively rare disease leading to small intestine obstruction. SEP leads to acute, subacute or chronic attacks of intestinal obstruction. Since it is a slow developing process, most patients have mild to severe intestinal obstruction signs and symptoms for a period of time. Although its aetiology is not clear, it is divided into primary (idiopathic) and secondary forms in which a trigger for the inflammatory process can be identified. Here, we present a patient with complete intestinal obstruction due to an abdominal cocoon.

Keywords: Intestinal obstruction, abdominal cocoon, rare cause of intestinal obstruction

INTRODUCTION

Abdominal cocoon was first described and named by Foo et al.[1] in 1978. Abdominal cocoon can be either primary (idiopathic) or secondary to an inflammatory process.[2] Primary SEP cases are mainly seen in adolescent women in tropical and subtropical areas, leading to theories that retrograde menstruation or gynaecologic infections are their causes.[1] However, there are other study results which state that the primary form of the disease can be seen at any age and in both sexes with a male to female ratio of 2:1.[3]

In the secondary form, various local or systemic factors can be identified as triggers of peritoneal inflammation. These factors may be medications (practolol, methotrexate, antiepileptic drugs), intra-abdominal infections (tuberculosis, bacterial peritonitis, cytomegalovirus, fungus, parasites), peritoneal dialysis, organ transplantations (liver, small intestine, kidney), intraperitoneal chemotherapy, foreign bodies, talcum powder, intraperitoneal iodine, asbestosis, silica, sarcoidosis, systemic lupus erythematosus, or familial mediterranean fever.[2]

CASE PRESENTATION

A 87-year-old male patient was admitted to the emergency department with abdominal pain, distension, vomiting and constipation for three days. He had a history of mild abdominal pain and distension attacks over the previous three years with spontaneous relief. He had undergone a right inguinal hernia operation 5 years ago in his medical history with no abdominal surgery, haemodialysis, primary peritonitis, tuberculosis, systemic organ disease or drug use. On physical examination, there was a mild abdominal distension with increased bowel sounds. On palpation, he had severe tenderness in the right upper and lower quadrants, but no rigidity or rebound sign. The patient's laboratory results were unremarkable except for rise in blood urea nitrogen and creatinine levels due to dehydration, with values of 130 mg/dL and 1.4 mg/dL, respectively. Plain abdominal X-ray revealed air fluid levels and dilated intestinal segments (Figure 1). Ultrasonography revealed dilated intestinal segments up to 4 cm in diameter in all quadrants of the abdomen. Abdominal computed tomography (CT) revealed diffuse dilated intestinal segments, and in the right lower quadrant, there was discontinuity of the intestinal segments, where there was a suspected

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obstruction with a conglomerate formation. The terminal ileum and the rest of the colonic segments were collapsed.

With the diagnosis of mechanical small bowel obstruction due yo unknown aetiology, the patient was hospitalized. Oral intake was stopped, intravenous fluid was started and a nasogastric tube was inserted. The nasogastric tube content was faecaloid. After 24 hours of observation, there was no clinical improvement in the patient's status. There was no regression in distension, no discharge of gas or faeces, and the nasogastric tube content was still faecaloid. The patient underwent an exploratory laparotomy on the second day of hospitalisation. During this exploration, small bowel conglomerate (abdominal cacoon) was found. The affected intestinal segment was between 200 cm from the ligament of Treitz and 30 cm proximal to the cecum. There were no other pathologic findings within the intra-abdominal cavity. The adhesions were dense and covered with pseudo capsules (Figure 2). Since this conglomerate was a compact structure, total excision was performed. After resection of the abdominal cocoon, a side to side small bowel anastomosis was performed using a linear stapler. The postoperative period was uneventful and he was discharged on the 10th postoperative day.

Written informed consent was obtained from the patient who participated in this study.

DISCUSSION

SEP is a rare cause of small bowel obstruction. Clinical presentations are non-specific; recurrent episodes of acute, subacute or chronic small bowel obstruction with abdominal distension, weight loss, nausea, vomiting, anorexia and vague abdominal pain are common in most cases.[4] Laboratory and radiologic findings are also non-specific. Therefore, the patients are usually diagnosed with a mechanical bowel obstruction. SEP diagnosis can be made perioperatively in most cases.

For preoperative diagnosis, plain abdominal X-ray, ultrasonography and/or CT scan can be used. Plain X-ray findings, which may show dilated loops of the small intestine with air-fluid levels, are not specific. Ultrasonography has been reported to facilitate the diagnosis and may reveal dilated bowel segments encased by a dense fibrous membrane.[5] Contrast-enhanced CT is the most useful imaging method for diagnosis of SEP. Small bowel loops, often tethered together in an enveloping peritoneum defined as the "cauliflower sign", is the characteristic appearance.[6] In a study on 22 patients with SEP, Gorsi et al.[7], found that the percentage of "cauliflower sign" was 64%. Despite all these imaging studies, preoperative diagnosis is still difficult.

SEP is classified as primary (idiopathic) or secondary, depending on the underlying pathology. Primary SEP can exist in three forms: type 1: partial capsulation of the small intestine; type 2: complete capsulation of the small intestine; and type 3: capsulation of the small and large intestine, ovary, liver, and stomach.[8] Although Primary SEP is idiopathic and is not associated with any certain cause, cytokines and fibroblasts likely influence the development of peritoneal fibrosis and neoangiogenesis.[9] In young girls living in tropical and subtropical regions, hypotheses including retrograde menstruation with a superimposed viral infection and retrograde peritonitis and cell-mediated immunologic tissue damage incited by gynaecological infections have been proposed.[9] However, these hypotheses do not explain the etiopathogenesis for all patients, since 75% of patients with primary SEP are men, premenstrual women or children, with a male to female ratio of 2:1.[9] Secondary SEP is more common, associated with many causes including prolonged beta blocker therapy (practolol), peritoneal dialysis, primary intra-abdominal infections, medications, organ transplantations, intraperitoneal chemotherapy, sarcoidosis, familial mediterranean fever, systemic lupus erythematosus, asbestosis, silica, foreign bodies, talcum powder and intra-abdominal iodine.[2]

SEP is a rare and usually slow progressive syndrome with recurrent episodes of small bowel obstruction signs and symptoms. Although some medical treatment modalities have been developed for those

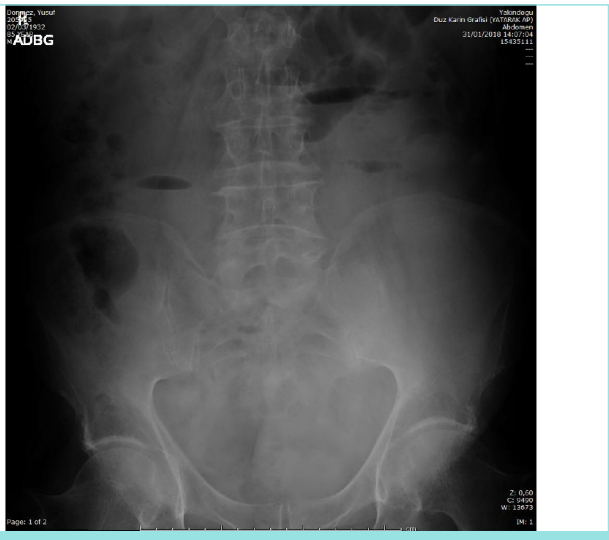

Figure 1. Air fluid levels and dilated intestinal segments on plain abdominal X-ray.

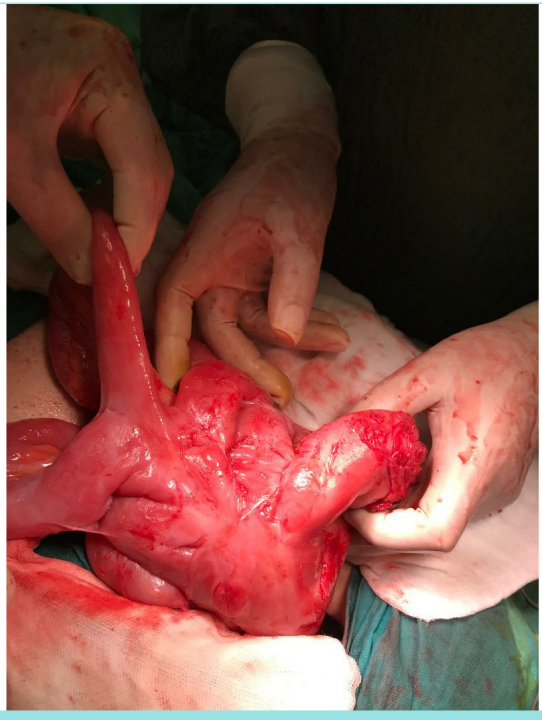

Figure 2. Small bowel conglomerate (abdominal cocoon) with dense fibrous pseudo capsule.

patients who are diagnosed before complete obstruction, especially in the Secondary SEP form, most patients need surgical intervention due to complete intestinal obstruction.[10] The preferred type of surgery is membrane excision and lysis of adhesions.[10] However, in some cases, resection of the affected segment is inevitable due to the dense adhesions. With awereness of this rare cause of intestinal obstruction, radiological imaging studies, especially CT scans, play a major role in establishing its diagnosis.

MAIN POINTS

- Abdominal cocoon is a rare and slow progressing syndrome with recurrent episodes of abdominal pain and distention.
- Laboratory and radiologic findings are usually non-specific.
- "Cauliflower sign" in CT is one of the most important imaging findings which may make preoperative diagnosis possible.
- Despite everything, definitive diagnosis can only be made perioperatively in most cases.

ETHICS

Informed Consent: Written informed consent was obtained from the patient who participated in this study.

Peer-review: Internally peer-reviewed.

Authorship Contributions

Concept: A.Ö., Design: A.Ö., Supervision: N.Ö., H.B., Materials: K.A., Data Collection and/or Processing: A.Ö., Analysis and/or Interpretation: K.A., Literature Search: A.Ö., Writing: A.Ö., Critical Review: N.Ö., H.B.

DISCLOSURES

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