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Idiopathic Encapsulating Peritonitis as Unusual Cause of Bowel Obstruction Discovered Intraoperative: A Case Report

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Abstract **Case Report**

Idiopathic sclerosing encapsulating peritonitis (SEP) is a rare cause of intestinal obstruction that is characterized by total or patial encasement of the intestines by a fibrocollagenous coccon-like a membrane. Early clinical features of this condition are generally non-specific and are frequently not recognized until the patient develops signs of small bowel obstruction. We report the observation of an adult patient operated for an acute bowel obstruction, in which the diagnosis of sclerosing encapsulating peritonitis (SEP) was made intraoperative. A good understanding of SEP and a better awareness of its clinical, paraclinical and therapeutic findings may aid to establish better management of this rare condition.

Keywords: Acute intestinal obstruction, sclerosing encapsulating peritonitis, small bowel, laparotomy.

INTRODUCTION

Sclerosing encapsulating peritonitis (SEP) is a rare cause of intestinal obstruction that is characterized by total or partial encasement of the intestines by a fibrocollagenous coccon-like a membrane [1]. A definite pre-operative diagnosis is difficult despite the contribution of modern imaging and this, due to the rarity and polymorphism clinical spectrum of this condition [2]. The etiology remains unknown and presumably is multifactorial. Treatment is also debated. We describe a case of an idiopathic encapsulating peritonitis discovered intraoperatively in a 51 year-old African male. Through this report case and a review of the literature, the authors emphazise the clinical features of this uncommon condition and discuss how to manage this kind of patients.

CASE PRESENTATION

A 51-year-old male Moroccan patient was presented in our hospital with a five days history of diffuse abdominal pain, which was associated with vomiting and inability to defecate or pass gas. The patient had no previous abdominal surgery, medication use or other medical history. The physical examination showed mild abdominal distension and hyperactive bowel sounds in pitch and frequency, but with non-tender. He was afebrile and hemodynamically stable. Findings of laboratory studies were a white blood cell count of 9600/ml and a C-reactive protein of 5,5 mg/l, Hemoglobin 12,3 g/dl, Blood urea 0,24 g/l and creatinine 8 mg/l, Kalemia 4,3 mmol/l and natremia 138 mmol/l. According to this symptomatology of the occlusive syndrome, a contrast-enhanced computed tomography (CT) scan was subsequently performed, which demonstrated generalized dilatation of the small bowel loops **(Fig. 1)**. Preoperative work-up did not reveal any specific etiology. Then, an exploratory median laparotomy was performed. At laparotomy, all small bowel segments were dilated up to the level of the terminal ileum. Terminal ileal loops were found covered with a thick membrane forming a sort of fibrous shell **(Fig. 2)**. The diagnosis of encapsulating peritonitis was made intraoperatively. Incision of the thick membrane and adhesiolysis of small bowel loops were performed **(Fig. 3)**. After the incision of the membrane, ileal loops were freed. Circulation of the bowel segment was intact; therefore, no resection was needed during the operation. The patient made an uneventful recovery and was discharged on the sixth postoperative day. In the successive follow-ups, the patient has been followed for five month post-operatively and is doing well.

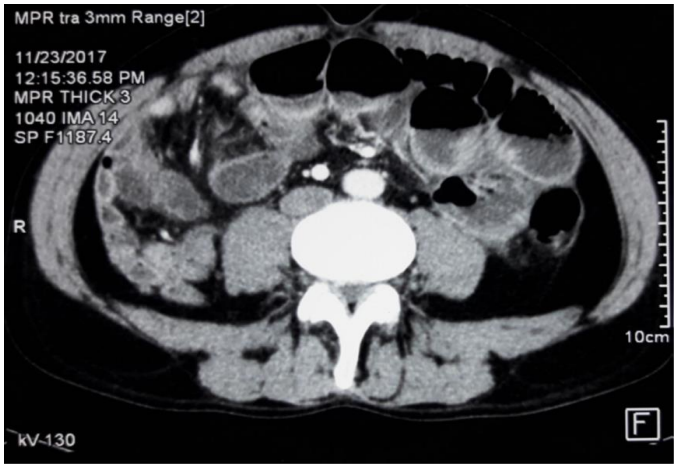

Fig-1: Computerized tomography (CT) scan of the abdomen showing dilated small bowel loops.

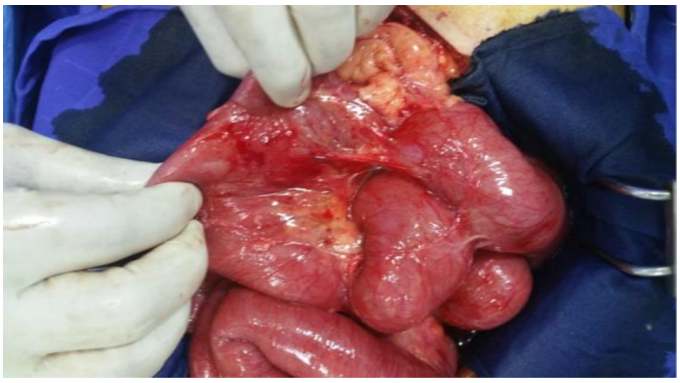

Fig-2: Intraoperative photography showing encapsulated of the small bowel in a thick membrane forming a fibrous shell.

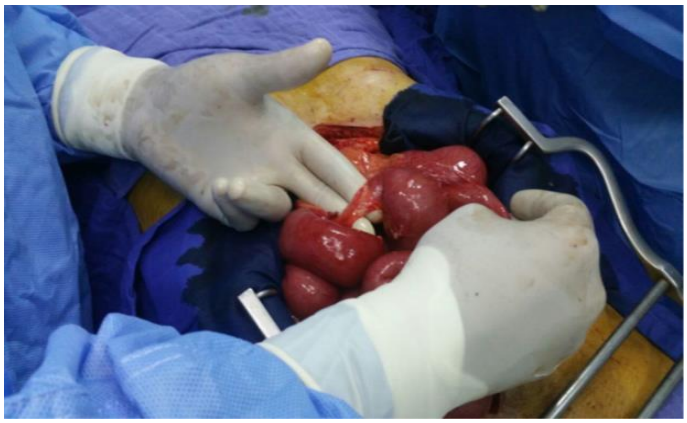

Fig-3: Intraoperative view after Incision of the thick membrane and adhesiolysis of small bowel loops.

Table-1: A comptemporary review of the literature regarding sclerosing encapsulating peritonitis.

	Li *et al*. [8] (n=65) 2014	**Wei *et al*.[1] (n=24) 2009**	**Singh *et al*.[11] (n=18) 2013**
Mean age diagnosis in years, (Range)	39(14-79)	48(16-70)	34(15-37)
Ratio Male/Female, n	57/8	9/15	8/10
Symptoms/Signs, n(%)	Abdominal pain 56(86,1) Abdominal distension 53(81,5) Nausea/vomiting 35(53,8) Abdominal mass 21(32,3) Fever 19(29,2)	Intestinal obstruction 11(45,8) Abdominal mass 10(41,7) Asymptomatic 3(12,5)	Abdominal pain 18(100) Abdominal mass 2(11,1) Tuberculosis abdominal 9(50)
Time diagnosis, n(%) Preoperative Operative	 31(47,7) 34(52,3)	 4(16,7) 20(83,3)	 -

Imaging modality	-	X-ray Scan CT	Scan CT
Treatment, n(%) Surgery Conservative approach	 65(100) 0	 24(100) 0	 17(94,5) 1(5,5)
Surgery type, n(%)	Excision+ adhesiolysis 42(64,6) Excision+adhesiolysis+intestinal stenting 23(35,3)	Excision+adhesiolysis 3(12,5) Excision+adhesiolysis+apendectomy 17(70,8) Excision+adhesiolysis+enterotomy 2(8,3) Excision+adhesiolysis+ceocofixation 2(8,3)	Excision+adhesiolysis 15(83,3) Excision+adhesiolysis+ileostomy 2(11,1)
Complications, n(%)	Recurrence obstruction 4(6,1)	Inflammatory bowel obstruction 3(12,5) Adhesive ileus 3(12,5)	Death due to liver failure 1(5,5)

Scan CT= Scan computerizd tomography.

DISCUSSION

The first documented case of EP was observed by Owtschinnikow in 1907, who labeled it peritonitis chronic fibrosa incapsulata, and the term sclerosing encapsulating peritonitis (SEP), also known as “idiopathic encapsulating peritoneal sclerosis” or “abdominal cocoon” was coined by Foo et al. in 1978 [3]. It results in the formation of a thick, white, pearly, fibrous membrane that completely or partially encasing the intestinal loops, leading to acute, subacute or chronic bowel obstruction [4].

Despite various hypotheses, the etiology is still unknown. SEP is divided into primary and secondary forms. The primary, or idiopathic form, is mostly seen in adolescent females living in tropical and subtropical regions such as those in the Indian subcontinent as well as China, Malaysia, Singapore, Nigeria, Kenya and South Africa. However, our patient was middle aged North-African male. As patients are generally asymptomatic and primary SEP is not obviously associated with other conditions. The typical age range is between 12 and 18 years; in fact, patient ages range from 6 to 79 years in the literature. However, the secondary form can be mainly secondary to chronic ambulatory peritoneal dialysis with an occurrence in around 0.9-7.3% of patients [5]. It has been associated with abdominal surgery, ventricle peritoneal shunts, retrograde menstruation, and tuberculosis peritonitis, the intake of practolol, sarcoidosis, carcinoid tumors or patients with liver cirrhosis. In patients with secondary SEP, a clinician may be alerted to the possible diagnosis due to predisposing factors such as parkinson’s disease, intraperitoneal shunts or autoimmune conditions.

The clinical presentation of patients with SEP is usually vague, with nonspecific abdominal symptoms, including bloating, colicky abdominal pain, constipation, nausea or vomiting, which is why most cases go undiagnosed for a long time [6]. It can also present with recurrent ascites or as an abdominal mass. Other patients, such as our patient, present with acute intestinal obstruction. In the literature, two cases of perforated bowel secondary to SEP in acute emergencies have been reported [7].

The preoperative diagnosis of primary SEP is usually challenging. In most patients, it is diagnosed by intraoperative findings and histopathological studies established during laparotomy which was the case in our patient. Li *et al*. found that 52.3% of a large series of 65 SEP patients were diagnosed during surgery in contrast to 47.7% who were diagnosed preoperatively [table 1]. Laparotomy allows confirmation of the diagnosis of EP and the realization of peritoneal biopsies that provide the diagnosis of certainty. Macroscopic examination reveals total or partial encapsulation of the small bowel, adherence of intestinal loops, fibrous thickening of the visceral peritoneum, and focal peritoneal bleeding. Although these are not pathognomonic to SEP, they usually support the diagnosis [2].

To properly diagnosis of EP preoperatively, imaging studies are of crucial importance. Radiographs may show diffuse air-fluid levels and dilatation of the small intestine in the form of loops. Barium studies usually show bowel loops that accumulate in the center of the abdomen, and this finding is called “sign of cauliflower or accordion”. CT scan is considered the most useful tool for diagnosing EP, especially multidetector CT with excellent image quality on coronal, sagittal, and axial planes. The characteristic CT sign is the appearance of loops of small intestine that conglomerate at midline and are encased by a dense mantle without peripheral contrast uptake. Additional findings may include peritoneal thickening, ascitis, intestinal obstruction, calcification of bowel wall, or lymphadenopathy [8, 9]. The findings of the magnetic resonnance imagery (MRI) enterography are usually similar to Scan CT.

There is no consensus regarding the optimal treatment in patients with SEP. However, evidence in the literature indicates that it is prudent to manage patients with minimal abdominal symptoms conservatively, with bowel rest, nasogastric

decompression and either enteral or parenteral nutritional support. Drug medical treatment may be also initiated for patients who fail to respond to conservative treatment, including tamoxifen, steroids, colchicine, azathioprine and mycophenolic acid. Some reports have described the effectiveness of these medications for patients with idiopathic SEP. In those patients with severe symptoms of intestinal obstruction and those who do not respond to conservative management may be candidates for surgical interventions.The treatment of choice for this condition is surgical, consisting of dissection of the membrane and extensive adhesiolysis [9]. Breaking of adhesions needs to be done carefully, to prevent damage to serosal surface and perforation. Bowel resection is indicated only when the intestine is nonviable. In our case, encapsulating membrane was excised and adhesions were released. Resection of bowel was not required, because bowel loops were not strangulated. In fact, early preoperative diagnosis and treatment of the EP is vital for the circulation of the bowel segments and for preventing the risk of strangulation. The administration of an antifibrogenic and anti-inflammatory agents during the postoperative period in patients in which the encapsulating membrane cannot be completely excised appears to be beneficial [1,10].

The prognosis is generally good, subject to proper care. But postoperative complications such as intestinal perforation may occur two to three days after enterolysis. The postoperative complication rate among patients who underwent enterolysis alone was 9.1% compared to 6.1% among those with internal splinting; moreover, the recurrence rate of intestinal obstruction was significantly higher [11].

CONCLUSION

Idiopathic encapsulating peritonitis (SEP) forms a minority of unusual conditions that lead to acute intestinal obstruction. Preoperative diagnosis is a true challenge for practitioners. A good understanding of SEP and a better awareness of its clinical, paraclinical and therapeutic findings may aid to establish better management of this rare condition.

Conflicts of interests

All authors of this article have not competing interests.

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The building and writing of this article has not fundings sources.

Consent

Written informed consent was obtained from the patient for the publication of this case report and its accompanying images.

Author's Contributions

Data collection: A. Houba, N. Doghmi; Analysis and data interpretation: A. Houba, N. Doghmi; Writing and revision of Article: A. Houba, N. Doghmi. All authors approved the final version of the manuscript.

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