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Sigmoido-Cecal Fistula: A Rare Case of Complicated Recurrent Diverticulitis and a Review of the Literature

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Patient: Female, 76
Final Diagnosis: Complicated recurrent diverticulitis
Symptoms: Abdominal pain • bloating • inability to pass stool and gas
Medication: —
Clinical Procedure: Hartmann's procedure
Specialty: Surgery

Objective: Unusual clinical course

Background: Although diverticular disease is well described and treated in daily clinical practice, there are cases that attract great interest because of their complexity and difficulty in management. Herein, we describe a rare case of colo-colonic fistula-complicated diverticulitis that necessitated urgent surgical intervention.

Case Report: A 76-year-old female patient with a known history of diverticular disease of the sigmoid colon presented in the Emergency Department for evaluation of left lower quadrant abdominal pain. The clinical and radiological examinations revealed a recurrent episode of acute diverticulitis of the sigmoid colon. However, it was of great interest that we detected a sigmoido-cecal fistula in the abdominal computed tomography (CT). The patient was admitted to the hospital for conservative treatment. After 48 hours, the patient's clinical status deteriorated, with pain aggravation, abdominal distension, bloating, and metallic bowel sounds. The simple abdominal x-ray revealed large-bowel obstruction and the CT demonstrated worsening inflammation of the sigmoid colon. An exploratory laparotomy revealed an inflamed dolichol-sigmoid colon forming a fistulous tract with the cecum and thus, mimicking a closed loop obstruction. The sigmoid colon was transected en bloc with the sigmoido-cecal fistula and a Hartmann's procedure was performed.

Conclusions: This case is extremely unusual as the patient presented at the same time two complications of diverticular disease, both obstruction and this rare formation of sigmoido-cecal fistula. It is presented in order to acquaint surgeons with the possibility of an unexpected course of this disease which indeed necessitates an individualized management.

MeSH Keywords: Cecum • Digestive System Fistula • Diverticulitis, Colonic • Intestinal Fistula • Sigmoid Diseases

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Background

Diverticular disease is nowadays very common in developed countries. Despite the evolving research, its pathogenesis still remains uncertain. Although the incidence of its associated complications is low, complications can be sometimes devastating. Complicated diverticulitis either by obstruction, fistula, abscess, or perforation is a surgical disease. Recent studies modified the guidelines concerning medical and surgical management, recommending that surgical treatment, either emergency or elective, should be determined in a case by case basis. Furthermore, elective surgery should be performed only in the presence of ongoing complications or in uncomplicated recurrent diverticulitis that affects a patient's quality of life [1].

Here, we discuss a very unusual case of a colo-colonic fistula-complicated diverticular disease. Although the formation specifically of sigmoido-cecal fistula is an extremely rare complication, it is a condition clinicians should be aware of. It is associated with a particular clinical manifestation and its treatment needs a special approach especially in an acute setting or when it coexists with other complications.

Case Report

A 76-year-old female patient with a known history of diverticular disease of the sigmoid colon presented in the Emergency Department for evaluation of left lower quadrant abdominal pain, which started 3 days ago. The patient denied any other accompanying symptoms. She described a few previous similar but less severe self-resolved episodes. She underwent a colonoscopy 3 years ago, revealing diverticular disease without obvious complications. Her medical and surgical history included atrial fibrillation, arterial hypertension, dyslipidemia, appendectomy, cholecystectomy, hysterectomy, and 2 cesarean sections. Her medication consisted of dabigatran, digoxin, carvedilol, and simvastatin.

At presentation, the patient was hemodynamically stable, afebrile, and the clinical examination showed diffuse left lower quadrant tenderness, with a positive rebound sign but no rigidity or guarding. Her white blood cell count was 8.43 per microliter with 68.5% neutrophils. C-reactive protein (CRP) was within normal limits. Because of her known history of diverticular disease, our first hypothesis was diverticulitis. An abdominal computed tomography (CT) scan with intravenous contrast was performed which demonstrated the presence of acute diverticulitis of sigmoid colon with colonic wall thickening and pericolic soft tissue changes (modified Hinchey classification Ia [2]). However, another finding drew our attention. A fistula between the sigmoid colon and the cecum was detected (Figure 1).

The patient was admitted to the hospital and conservative treatment with intravenous (IV) fluids, IV antibiotics (ciprofloxacin 400 mg ×2 IV, metronidazole 500 mg ×3 IV) and bowel rest was initiated. A colonoscopy and further investigation for the

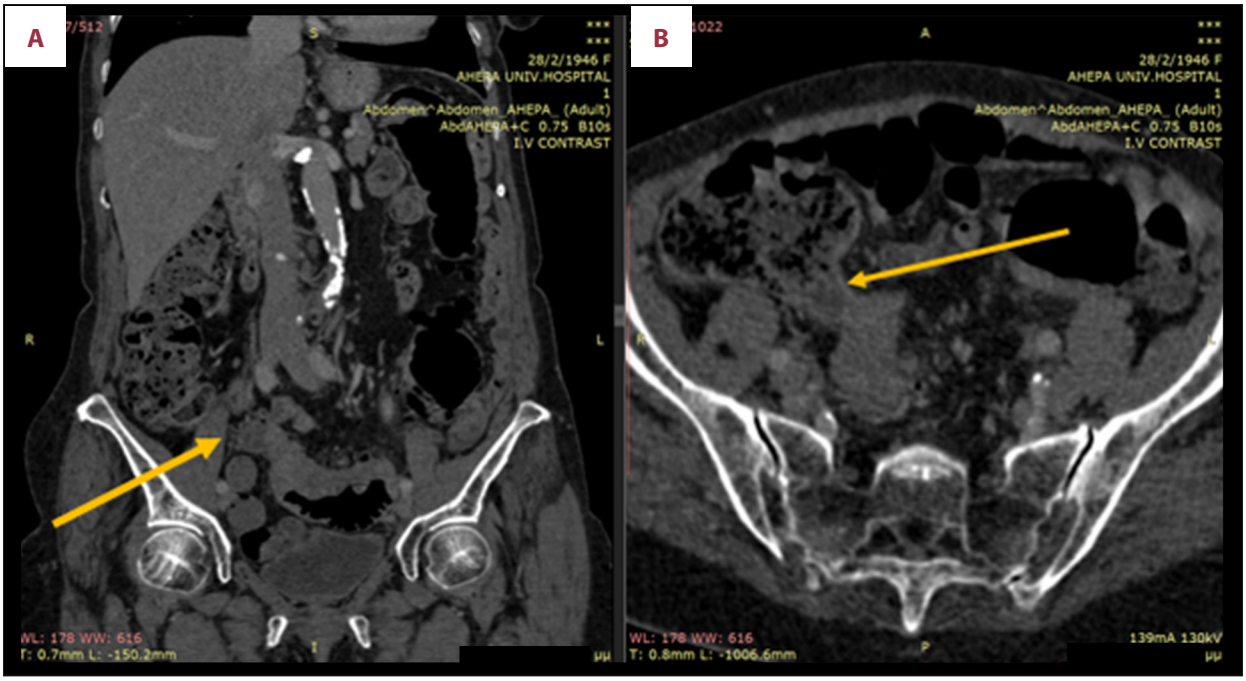

Figure 1. Computed tomography scan at admission (**A, B**), detection of the fistulous tract (arrows) between the sigmoid colon and the cecum.

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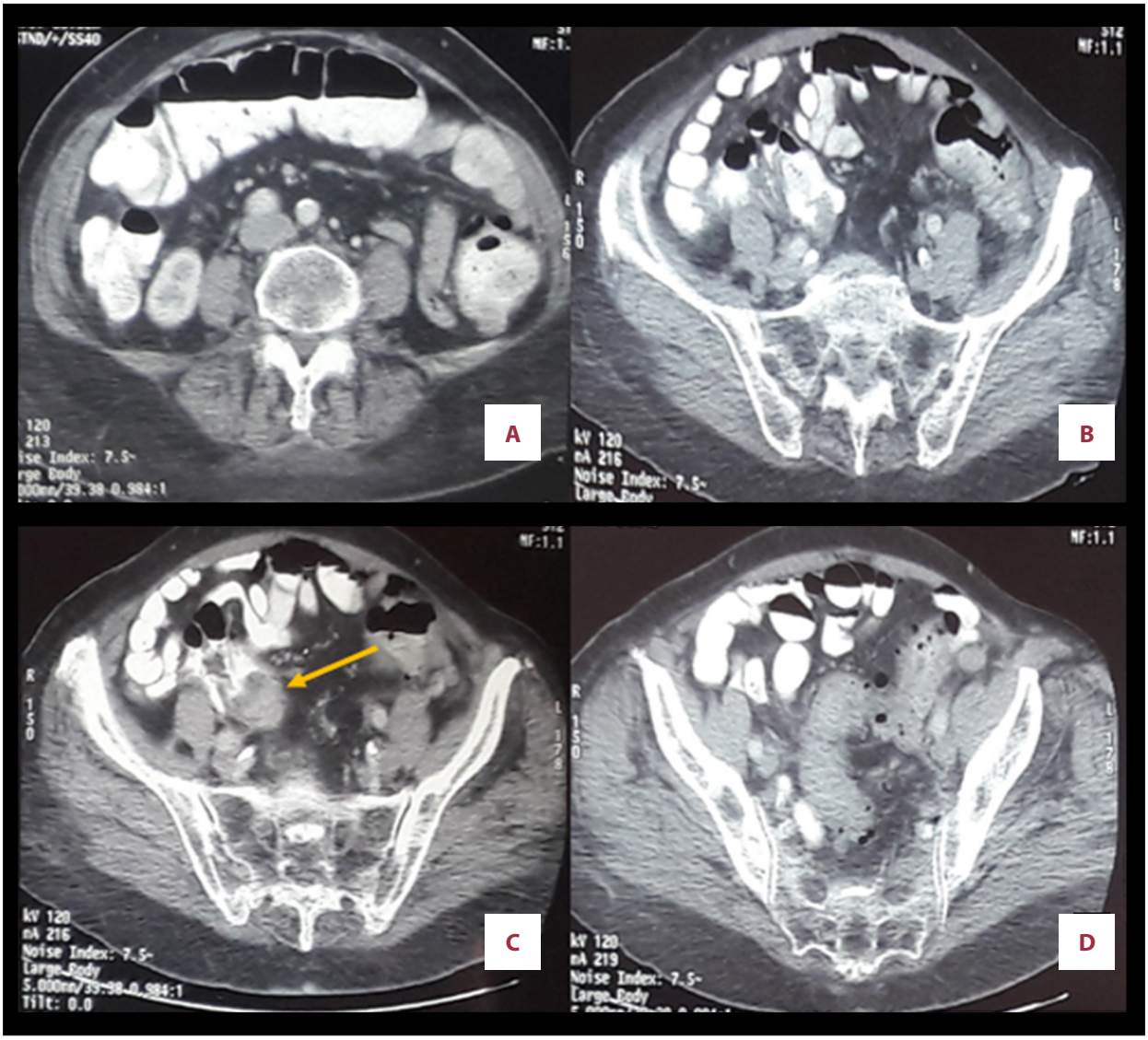

Figure 2. Computed tomography scan after deterioration of clinical status (**A, B, D**) large bowel obstruction due to diverticulitis of the sigmoid colon (**C**) filling defect in the cecum (arrow).

colo-colonic fistula was planned after 6 to 8 weeks. An elective surgical approach after this complicated recurrent episode was decided.

However, 2 days after admission, the patient complained about pain aggravation, nausea, and inability to pass stool and gas. Pertinent findings on physical examination included abdominal distension with bloating and metallic bowel sounds. The abdominal palpation revealed a moderate tenderness with rebound in the left lower abdominal quadrant, which was at this time extended at the epigastric and hypogastric region. There was only a mildly elevated CRP in blood examinations.

We hypothesized that this episode of acute diverticulitis was twice complicated and apart from the fistula formation, in a background of colonic stricture, probably resulted to obstruction. A simple abdominal x-ray confirmed our suspicion revealing large-bowel obstruction. The patient underwent an abdominal CT scan with intravenous and oral contrast, which demonstrated the large bowel obstruction and worsening inflammation of the sigmoid colon. The fistulous tract was now illustrated as a filling defect in the cecum (Figure 2). Because of this deterioration of both clinical and imaging status, the patient was taken to the operating room.

An exploratory laparotomy was performed and revealed a large-bowel obstruction with an intensively distended colon in its entity, from the proximity of the sigmoid colon up to the cecum, mimicking a closed loop obstruction. A complicated diverticulitis of the sigmoid colon which probably led to a total

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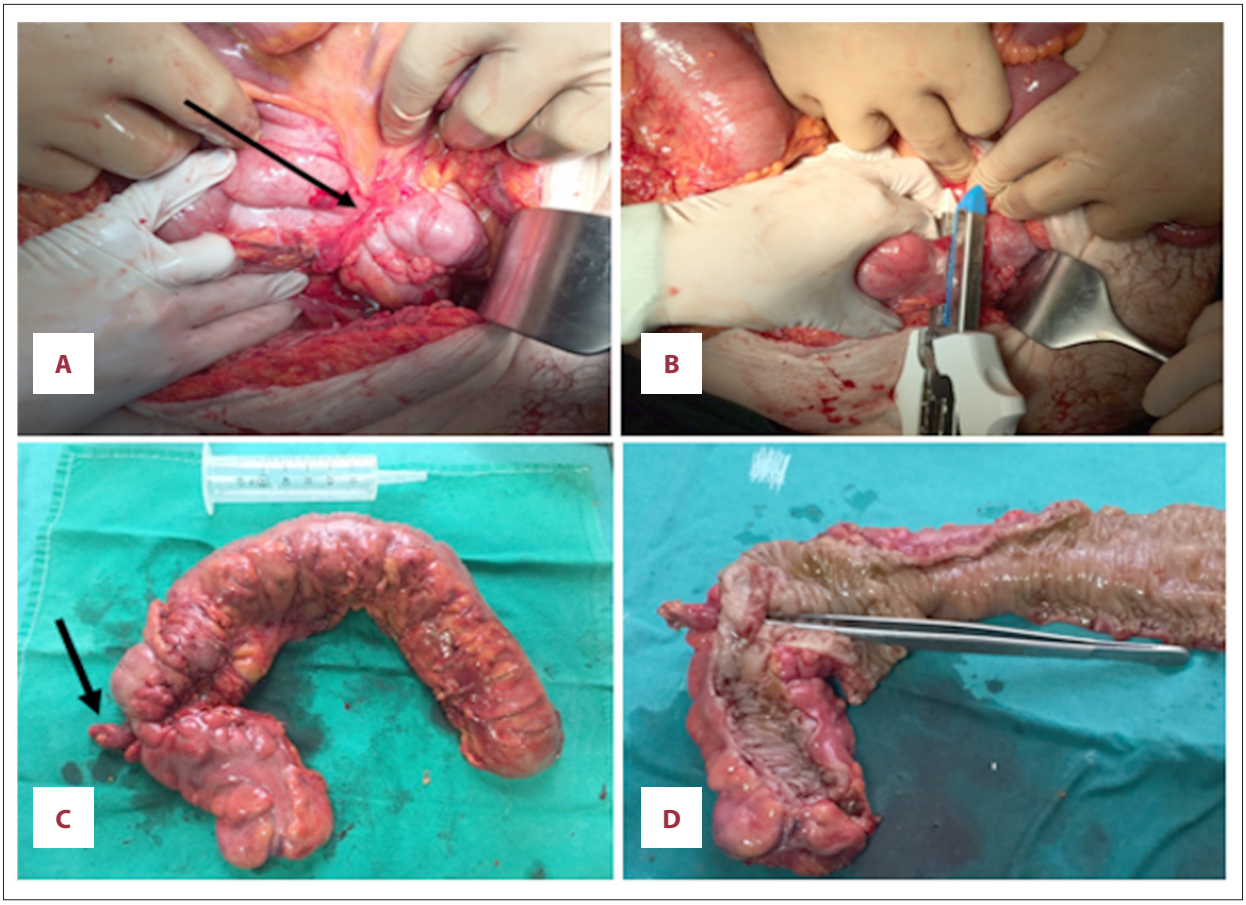

Figure 3. Intraoperative findings: (**A**) sigmoido-cecal fistula, (**B**) excision of the sigmoido-cecal fistula with a part of healthy cecal wall with the use of a linear cutting stapler. Colonic specimen (**C, D**): sigmoid colon en bloc with the sigmoido-cecal fistula.

obstruction was the cause of this condition. The decision to perform a Hartmann's procedure was made. Intraoperatively, we observed an inflamed dolichol-sigmoid colon in close proximity to the cecum. The mobilization of the redundant sigmoid colon revealed the colo-colonic fistula, probably between a ruptured diverticula of the sigmoid colon towards the cecum, corresponding to the imaging finding described in the CT scan.

A typical Hartman's procedure was performed. The sigmoid colon was completely mobilized and transected en bloc with the sigmoido-cecal fistula. The sigmoido-cecal fistula with a part of healthy cecal wall was excised using a linear cutting stapler (Figure 3).

A drain was placed in the right paracolic gutter ending in the pouch of Douglas, thus including also the drainage at the site of fistula excision from the cecum, in order to control and be aware of any leak. The surgical procedure was uneventful and lasted approximately 1 hour and 30 min.

The patient resumed oral intake on postoperative day 2 and the drainage catheter was removed on postoperative day 5. Hospitalization was uneventful, and the patient was discharged home on postoperative day 6.

The histopathologic examination revealed multiple diverticula reaching the adjacent fibrofatty tissue, all along the colonic specimen and chronic inflammation resulting in bowel wall thickening and fibrosis. A fistulous tract was detected, deriving from the sigmoid colon. Twenty-three lymph nodes were dissected with characteristics of inflammatory reactive lymphadenopathy. No malignancy was detected.

The patient underwent a restoration of the intestinal continuity after 3 months, which was uneventful. She has follow-up regularly with a gastroenterologist. She exhibits good health and complains of no discomfort.

Discussion

The aim of this report was to highlight that although diverticular disease is a well described and studied entity, it may occasionally present in an unusual and extraordinary way. Therefore,

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it is of great importance that the presence of a very unusual complication of the disease, the formation of a sigmoido-cecal fistula, which is rarely described in literature, should be considered. It was of great interest that this patient presented at the same time with 2 complications of diverticular disease, both obstruction and fistula formation, resulting in this particular clinical manifestation.

Diverticular disease is very common in western countries. Taking under consideration the rise in life expectancy and the concomitant increase of the incidence of this disease with aging, diverticulosis is a healthcare system burden. Although 80% to 85% of patients with diverticulosis are asymptomatic, approximately 15% will present symptomatic diverticulosis (diverticular disease) but without inflammation and 10% to 20% will suffer from acute diverticulitis [3].

The natural history of this disease varies, as it might remain asymptomatic, give rise to a single index episode, or become complicated or recurrent. The incidence of recurrent diverticulitis is estimated to 13% to 23% while the risk of recurrence increases after the second and third episode [3,4]. Complicated diverticular disease accounts about 25% of cases, characterized by either abcesses or fistula formation, obstruction, peritonitis and sepsis [5]. Contrary to what would be expected, complications more often occur during the first episode and recurrent diverticulitis does not predispose to complications [6].

Diverticular disease might be complicated in many ways and patients can present with more than one complication either simultaneously or in temporally separated episodes. In a study by Bharucha et al., 71% of patients had only 1 complication of their disease, and 22% and 7% had 2 and 3 or more complications, respectively. More than half of these patients suffered from simultaneous complications [6]. In our case, this female patient had 2 simultaneous complications of diverticulitis, both obstruction and fistula. There was no evidence to determine if the fistula or the partial obstruction occurred first, or if there was a causal link between them. Both of these complications have been thoroughly but separately investigated and described. It is well established that the inflammatory background of diverticular disease leads to these types of complications, both for partial/total obstruction and fistulation. However, we don't know if during their coexistence, one acted as an additional risk factor, along with diverticulitis itself, for the other complication. A logical hypothesis is that our patient who suffered from recurrent diverticular disease probably had developed colonic tissue alterations due to chronic inflammation resulting in fibrosis, stricture, and obstruction. Under acute circumstances, such as during a recurrent episode, the inflammatory process could lead to worsening of the already existed fibrosis and stricture, partial or even total obstruction, causing increase in intraluminal pressure. Together with the exacerbation of the disease, this could more easily result in the rupture of a diverticulum towards an adjacent organ and formation of a fistulous tract. It would be of great value to clarify if the course of each complication was independent of the other or if there was a causal aggravating link between them, as this would result in a better treatment planning.

Although fistula formation complicating diverticulitis is often described in the literature, colo-colonic fistulas are indeed very rare. Fistulas occur in 2% of patients with complicated diverticular disease, more commonly as colovesical (65%) and colovaginal (25%) fistulas [5], but they can involve any organ in the pelvis at unusual sites such as the uterus, the fallopian tube, the perineum, the ureter and as in our case, other parts of colon. Clinicians should also be aware of the possible existence of multiple fistulas, which can be either external or internal. Although rare, 3-way fistulas may also occur, involving colon, uterus, and bladder or colon, small bowel, and vagina [7]. Multiple tracts are found in 8% of patients [5].

Colo-colonic fistulas were first reported by Mayo and Blunt in 1950 in a series of 202 patients with diverticulitis, where 3 colo-colonic fistulas were detected [8]. Colcock and Stathamm treated 42 patients with fistulas complicating diverticular disease of the sigmoid colon and identified only 1 patient with a colo-colonic fistula, formed between the sigmoid and the descending colon [7]. In the aforementioned studies, it was stressed out that this kind of fistulas occurring between the sigmoid colon and any other part of the colon usually co-exists with multiple internal fistulas.

Sanowski and Costello described for the first time a case of sigmoido-cecal fistula as an unusual complication of acute diverticulitis [9]. In a review of the literature, we found only 4 cases of sigmoido-cecal fistulas [9–12], and an additional case which was malignant [13]. Almost all of the aforementioned reports referred to specific aspects of this rare complication.

Ramos et al. emphasized a possible impact (side effect) of nicorandil, a potassium channel opener with a nitrate component used in the management of angina, in the pathogenesis of diverticular sigmoido-cecal fistula [12]. On the other hand, Chung et al. suggested that the presence of a sigmoido-cecal fistula in complicated diverticular disease might result in obstruction but in a different way than we have described. According to their case report, a sigmoido-cecal fistula caused bacterial overgrowth and led to colonic pseudo-obstruction in a patient with diverticular disease, but no evidence of diverticulitis [11]. In contrast, in our case, the patient suffered from a recurrent episode of diverticulitis, which led to a real obstruction. We believe that the presence of the sigmoido-cecal fistula played a crucial role in our patient's clinical deterioration. The inflamed sigmoid colon, being fixed

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to the cecum because of the fistula formation, resulted in the worsening of the obstruction. Furthermore, the fistulous tract allowed a continuous drainage of enteric contents from cecum towards the sigmoid colon. This situation, in combination with the sigmoid obstruction, simulated a closed loop structure.

Finally, Hyun et al. referred to the diagnostic tools that are helpful in this uncommon complication. They recommended radiologic studies with contrast media, abdominal CT and colonoscopy combined with chromoendoscopy [10]. It is true that all of the aforementioned modalities can be useful in the diagnosis of sigmoido-cecal fistula, depending, however, on the clinical status of the patient and the manifestation of her or his disease. Under acute circumstances, as in our case, CT scan seems to be the most appropriate imaging technique, also revealing the fistulous tract.

The evolving research on diverticular disease modifies the treatment strategies considering each patient's profile. The decision for a surgical approach today is made on a patient by patient basis. To patients with symptomatic complicated diverticular disease (fistula and stenosis) elective or semi-elective surgery is recommended for symptomatic relief [4,14,15]. However, we should be aware that approximately 15% to 30% of patients admitted for the management of diverticulitis will require an urgent surgical intervention during admission, with an associated mortality rate of 18% [5]. A subset of these patients will just not respond to non-operative treatment, as for example in cases of infection-related ileus or bowel obstruction, as was found in our case. Our patient was taken urgently to the operating room because of the clinical and imaging findings, in a setting of individualized patient care approach. Hartmann's procedure remains a gold standard in the treatment of acute complicated diverticulitis in the emergency setting. Although laparoscopic approaches, even in diverticular fistulas, begin to gain acceptance [3], that approach was not feasible in our case because of the fistula location and the need for excessive resections. However, taking advantage of the confined inflammation to sigmoid colon, leaving cecal tissue healthy and intact, we were able to use a minimal invasive approach with the use of a linear cutting stapler in the cecal side of the fistula.

Conclusions

Although diverticular disease is often presented in a typical manner and its course is more or less predictable, it might occasionally surprise healthcare providers. Our case presented a paradigm of a complicated recurrent episode of diverticulitis with a very interesting clinical manifestation because of the extraordinary complication of a sigmoido-cecal fistula formation in combination with large bowel obstruction. Healthcare providers should be aware of this rare type of fistula formation and also aware of the possibility of concurrent complications.

Based on recent studies, there is a trend to individualize the management of diverticular disease on a patient by patient basis. Both the aforementioned situations (fistula and obstruction) require a surgical treatment in either elective or semi-elective setting [1]. In conclusion, it is our strong belief that surgical management of these complications shouldn't be delayed, in order to avoid any similar situations. In the acute and emergency setting of our case, taking also under consideration the presence of the sigmoido-cecal fistula, the large bowel obstruction and the patient's condition, we believe that Hartmann's procedure was the ideal surgical procedure, lacking risks like insecure anastomosis or excessive long-lasting manipulations and resections. On the other hand, in an elective setting, it is the surgeon's decision which of the recommended single or multiple staged, open or laparoscopic resection procedures he will perform. Concerning the surgical treatment of the sigmoido-cecal fistula, based on the principles of management of enteric fistulas, the resection of the fistulous tract with re-establishment of the gastrointestinal tract is strongly recommended [16]. In our case, taking advantage of the healthy cecal wall, the use of a linear cutting stapler was a great option, this way avoiding cecal resection.

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