



Contents lists available at ScienceDirect

## International Journal of Surgery Case Reports

journal homepage: [www.casereports.com](http://www.casereports.com)

## Presentation with perforation of the terminal ileum and acute limb ischemia in Crohn's disease: A case report

Anis Haddad<sup>a,c</sup>, Ahmed Ben Mahmoud<sup>a,c,\*</sup>, Youssef Chaker<sup>a,c</sup>, Alia Zehani<sup>b,c</sup>, Rachid Ksantini<sup>a,c</sup>, Montasser Jameleddine Kacem<sup>a,c</sup>

<sup>a</sup> Department of Surgery A, Rabta Hospital, Tunis, Tunisia

<sup>b</sup> Department of Pathology, Rabta Hospital, Tunis, Tunisia

<sup>c</sup> Faculty of Medicine of Tunis, Tunis El Manar University, Tunis, Tunisia



### ARTICLE INFO

#### Article history:

Received 19 January 2021

Received in revised form 3 February 2021

Accepted 4 February 2021

Available online 9 February 2021

#### Keywords:

Crohn's disease

Fecal peritonitis

Free peritoneum perforation

Limb acute ischemia

### ABSTRACT

**INTRODUCTION AND IMPORTANCE:** Crohn's disease is a chronic inflammatory bowel disease with complex pathophysiology and multiple complications, some of which can be fatal. We report herein the management of an unusual case of Crohn's disease revealed by two life-threatening complications.

**CASE PRESENTATION:** A 59-year-old patient presented with an acute abdominal pain evolving for one day with a clinical presentation in favor of peritonitis by perforation of the last ileal loop and acute limb ischemia.

At emergency laparotomy, we found a fecal peritonitis by perforation of the last ileal loop. The patient underwent an ileo-caecal resection with rifle barrel ileo-colostomy associated with embolectomy using a Fogarty catheter of the femoral artery. Pathological examination of the specimen showed an aspect consistent with an ileo-caecal Crohn's disease and blood clot embolus of the femoral artery. Postoperative course was uneventful and the patient was kept in remission with immunosuppressants.

**DISCUSSION:** Several complications may arise during the evolution of the disease. However, Life-threatening complication scarcely inaugurate crohn's disease like in our patient. Free perforation of the small intestine in crohn's disease occurs rarely, which makes its statistical study difficult. Risk factors for perforation are still poorly identified. Patients diagnosed with crohn's disease have a higher risk of thromboembolic complications. Indeed, there is an association between the activity of the disease and thromboembolic events. In our case, the severity of the clinical presentation as well as its inaugural character are unique.

**CONCLUSION:** The management of inaugural two uncommon acute conditions in Crohn's disease is challenging.

© 2021 The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

## 1. Introduction

Crohn's disease is an inflammatory disease of the digestive tract. The etiopathogenesis remains uncertain. This gastrointestinal disorder is responsible for transmural lesions. Various complications can inaugurate the disease such as stenosis, fistula, abscess and perforation in the free peritoneum.

We describe herein a rare case of Crohn's disease revealed by two life-threatening complications consisting in a fecal peritonitis by stercoral perforation of the last ileal loop associated with an acute ischemia of the lower limb in a 59-year-old-male patient.

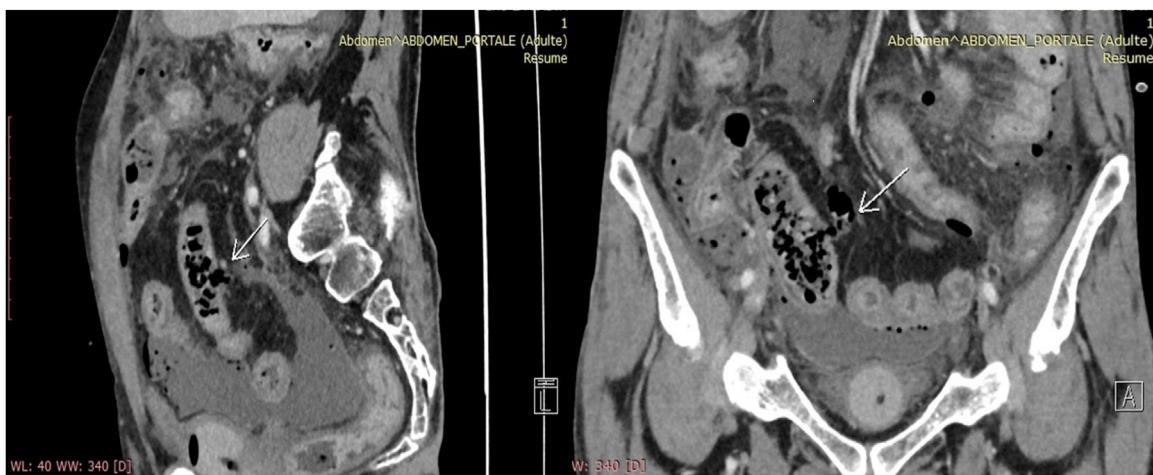
The case report has been reported in line with the SCARE criteria [1].

## 2. Case report

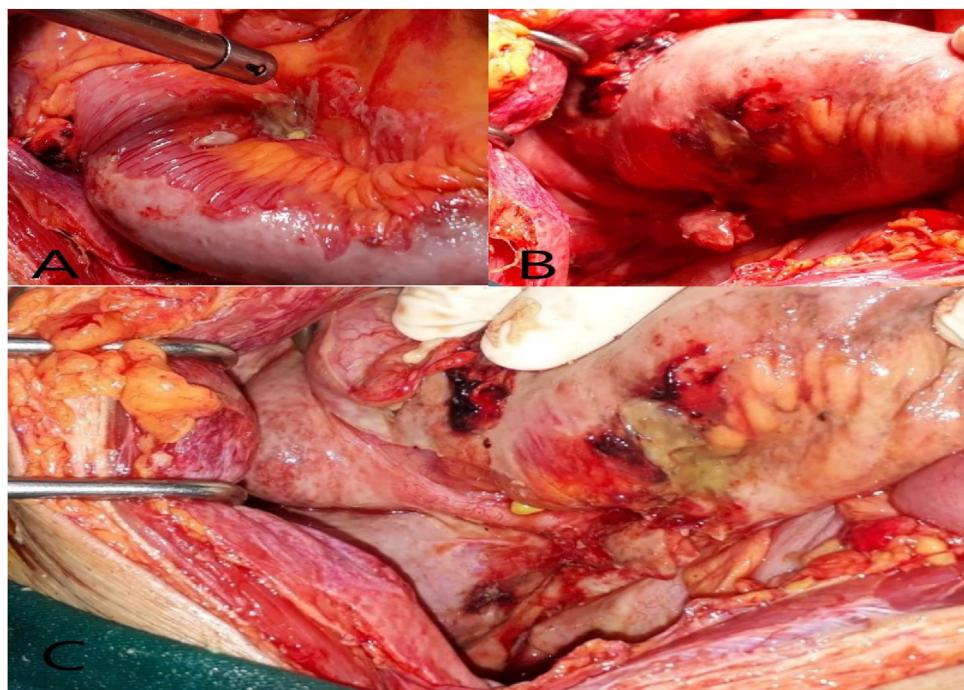
A 59-year-old patient with no past medical or surgical history, presented on the 31st of July 2018 to our emergency department with an acute diffuse vague abdominal pain evolving for less than 24 h associated with fever and incoercible vomiting. The patient did not report any past bowel disorders or recent general condition impairment. On physical examination, we found fever (38.5 °C) as well as a general abdominal guarding. Furthermore, the patient had left lower limb cooling with abolition of the popliteal pulse consistent with an acute ischemia. The hemodynamic state was stable and the respiratory rate was normal. Laboratory tests revealed a significant biological inflammatory syndrome with high white blood cell count (16,500/ $\mu$ l) and C-reactive protein level (278 mg/l).

\* Corresponding author at: Department of Surgery "A", La Rabta Hospital, La Rabta Jebbari, 1007, Tunis, Tunisia.

E-mail address: [ahmed.benmahmoud92@gmail.com](mailto:ahmed.benmahmoud92@gmail.com) (A. Ben Mahmoud).



**Fig. 1.** Parietal ileal defect with peritoneal effusion and pneumoperitoneum (white arrows).



**Fig. 2. (A, B and C):** Sclerolipomatosis, perforation of the last loop and advanced peritonitis with false membranes.

Consequently, we performed a chest X-Ray that showed a pneumoperitoneum of great abundance, suggesting the perforation of an intra-abdominal hollow organ.

An abdominal CT was therefore essential (Fig. 1). It showed a pneumoperitoneum and an intraperitoneal effusion of great abundance as well as a diffuse thickening enhancement of the peritoneal sheets. The ileal loops were distended associated with regular circumferential parietal thickening and submucosal edema. Significant sclerolipomatosis and comb sign were additionally noticed. Bone window study also revealed bilateral sacroiliitis. The rest of the exam showed no abnormalities.

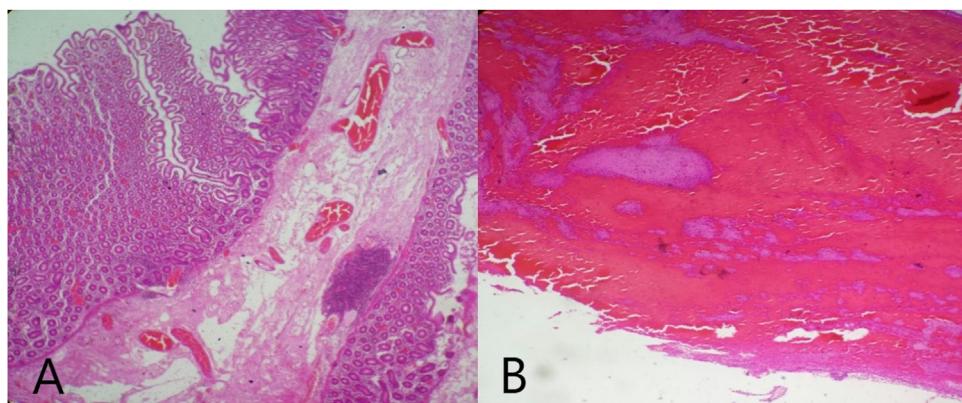
We performed an emergency surgery on the 1st of August 2018 through a median incision, after fluid and electrolytes resuscitation. On the exploration of the abdominal cavity, we found a fecal peritonitis with abundant intraperitoneal effusion and false membranes due to a punch perforation in the last ileal loop. No tumor-like thickening next to the perforation was noticed.

Important mesenteric sclerolipomatosis was suggestive of Crohn's disease (Fig. 2).

We performed an ileo-caecal resection with rifle barrel ileocolostomy in right iliac associated with abundant peritoneal toilet. Additionally, a Fogarty catheter was used to perform an embolotomy of the femoral artery with extraction of a blood clot.

Pathology examination of the specimen Fig. 3 showed chronic granulomatous ileitis lesions complicated by peritonitis which is consistent with Crohn's disease without vasculitis lesions. Histology of the embolus showed a bloody material.

The postoperative course was uneventful and the patient was put on heparin then vitamin K antagonists. He was discharged on Day 10. He was referred to gastroenterology department in order to initiate medical treatment for crohn's disease. Three months later, we performed an ileocolic lateral anastomosis. The patient was kept in clinical and endoscopic remission under immunosuppressants for the last two years.

**Fig. 3.** Pathology findings of the specimen.

A: Architectural disorganization of the ileal mucosa with a type of elongation and distortion of the crypts.

B: Ileal mucosa largely ulcerated covered with a fibrino-leukocytic coating.

### 3. Discussion

Crohn's disease is a chronic bowel condition that might result from a combination of genetic susceptibility, environmental factors and altered gut microbiota. Typically, this disorder of the gastrointestinal tract occurs in young people presenting with abdominal pain, chronic diarrhea and progressive impairment of the general state, evolving in a relapsing and remitting manner [2]. Several complications may arise during the evolution of the disease. However, life-threatening complication scarcely inaugurate Crohn's disease like in our patient.

The diagnosis of peritonitis is easily established on physical examination. Otherwise, etiological research remains a real challenge especially in elderly patient with no past history of inflammatory bowel disease, as we reported in our case. For this reason, CT-scan is compulsory to suspect the diagnosis and the complication efficiently [3]. The only evocative radiological feature in our patient was sclerolipomatosis.

Free perforation of the small intestine in crohn's disease occurs in less than 3 % [4,5]. The cases of Crohn's disease inaugurated by peritonitis are described in the 80 s and 90 s period [6] when the pathophysiology of the disease was not clear as well as the diagnostic and therapeutic means.

However, the Connect Study reported a high incidence of free perforation in Crohn's disease (6.5 %) compared to worldwide incidence [7]. This result was similar to those reported by a Japanese meta-analysis [8], suggesting inflammatory bowel disease phenotypes differences between Asian and Caucasian population. Perforation of the last ileal loop initiating Crohn's disease is a rare entity, which makes its statistical study difficult. A Korean study demonstrated also that among 88 patients, the perforation was the presenting sign in 46 patients (52 %) [7].

Ileal perforation was significantly high among the Korean (86.2 %) and Japanese (80 %) populations comparatively to Greenstein et al. results (50 %) [7,9].

Risk factors for perforation are still poorly identified. They may be related to the complex pathophysiology of the disease associating transmural lesions with very significant inflammatory activity and ischemia or to abscess with secondary perforation. The perforation may be due to a very significant distension upstream of a stenosis [9,10]. In our case, the first hypothesis is the most probable. Kim et al. reported that anti TNF therapy could be a risk factor for perforation [11]. Our patient was TNF treatment-naïve.

The perforation of the last ileal loop complicated by peritonitis remains serious and potentially fatal and the treatment of choice is an ileocaecal resection with or without an ileocolostomy [5,8,12].

Patients diagnosed with Crohn's disease have a higher risk of thromboembolic complications. Indeed, there is an association between the activity of the disease and thromboembolic events [13]. The incidence of thromboembolic events in inflammatory bowel diseases ranges from 1 to 8 % [14]. Surgery, immobilization, dehydration and the use of central venous catheter are risk factors related to the disease itself. In our case the severity of the thromboembolic accident as well as its presenting character are unique.

### 4. Conclusion

Crohn's disease remains a frequent pathology with variable diagnostic and therapeutic means, thereby reducing the incidence of inaugural complications through serious complications that can jeopardize the vital prognosis.

#### Declaration of Competing Interest

The authors report no declarations of interest.

#### Funding

None.

#### Ethical approval

Not applicable.

#### Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

#### Registration of research studies

Not Applicable.

**Guarantor**

Ahmed Ben Mahmoud.

**Provenance and peer review**

Not commissioned, externally peer-reviewed.

**CRediT authorship contribution statement**

**Anis Haddad:** Writing - original draft. **Ahmed Ben Mahmoud:** Writing - original draft. **Youssef Chaker:** Supervision. **Alia Zehani:** Data curation. **Rachid Ksantini:** Supervision. **Montasser Jameled-dine Kacem:** Supervision, Data curation.

**References**

- [1] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, A. Thoma, A.J. Beamish, A. Noureldin, A. Rao, B. Vasudevan, B. Challacombe, B. Perakath, B. Kirshtein, B. Ekser, C.S. Pramesh, D.M. Laskin, D. Machado-Aranda, D. Miguel, D. Pagano, F.H. Millham, G. Roy, H. Kadioglu, I.J. Nixon, I. Mukhejee, J.A. McCaul, J. Chi-Yong Ngu, J. Albrecht, J.G. Rivas, K. Ravendran, L. Derbyshire, M.H. Ather, M.A. Thorat, M. Valmasoni, M. Bashashati, M. Chalkoo, N.Z. Teo, N. Raison, O.J. Muensterer, P.J. Bradley, P. Goel, P.S. Pai, R.Y. Afifi, R.D. Rosin, R. Coppola, R. Klappenbach, R. Wynn, R.L. De Wilde, S. Surani, S. Giordano, S. Massarut, S.G. Raja, S. Basu, S.A. Enam, T.G. Manning, T. Cross, V.K. Karanth, V. Kasivisvanathan, Z. Mei, The SCARE 2020 Guideline: updating Consensus Surgical CAse REport (SCARE) Guidelines, Int. J. Surg. (2020), <http://dx.doi.org/10.1016/j.ijssu.2020.10.034>.
- [2] J. Torres, S. Mehandru, J.F. Colombel, L. Peyrin-Biroulet, Crohn's disease, Lancet 389 (2017) 1741–1755, [http://dx.doi.org/10.1016/S0140-6736\(16\)31711-1](http://dx.doi.org/10.1016/S0140-6736(16)31711-1).
- [3] A.J. Greenup, B. Bressler, G. Rosenfeld, Medical imaging in small bowel Crohn's disease - Computer tomography enterography, magnetic resonance enterography, and ultrasound: "Which one is the best for what", Inflamm. Bowel Dis. 22 (2016) 1246–1261, <http://dx.doi.org/10.1097/MIB.0000000000000727>.
- [4] S. Katz, N. Schulman, L. Levin, Free perforation in Crohn's disease: a report of 33 cases and review of literature, Am. J. Gastroenterol. 81 (1986) 38–43.
- [5] H.J. Freeman, Spontaneous free perforation of the small intestine in adults, World J. Gastroenterol. (2014) 9990–9997, <http://dx.doi.org/10.3748/wjg.v20.i29.9990>.
- [6] P.P. Casteleyn, J.C. Pector, C. Melon, Acute free perforation as first sign of Crohn's disease, Acta Chir. Belg. 77 (1978) 181–186.
- [7] J.W.K. Young Seok Doh, You Sun Kim, Song I. Bae, JongPil Im, JaeHee Cheon, ByongDuk Ye, D.S.H, W.H.K. Young Sook Park, Ji Hyun Lee, Young-Ho Kim, JooSung Kim, The clinical characteristics of patients with free perforation in Korean Crohn's disease: results from the CONNECT study, BMC Gastroenterol. 205 (2015) 72–76, <http://dx.doi.org/10.1186/s12876-015-0262-x>.
- [8] H. Ikeuchi, T. Yamamura, Free perforation in Crohn's disease : review of the Japanese literature, J. Gastroenterol. (2002) 1020–1027.
- [9] J.A.J. Greenstein, D.B. Sachar, D. Mann, P. Lachman, T. Heimann, and A H Aufses, greenstein1987.pdf, Ann. Surg. 205 (1987) 72–76.
- [10] M. Gajendran, P. Loganathan, A.P. Catinella, J.G. Hashash, A comprehensive review and update on Crohn's disease, Disease-a-Month 64 (2018) 20–57, <http://dx.doi.org/10.1016/j.dismonth.2017.07.001>.
- [11] J.W. Kim, H.S. Lee, B.D. Ye, S.K. Yang, S.W. Hwang, S.H. Park, D.H. Yang, K.J. Kim, J.S. Byeon, S.J. Myung, Y.S. Yoon, C.S. Yu, J.H. Kim, Incidence of and risk factors for free bowel perforation in patients with Crohn's disease, Dig. Dis. Sci. 62 (2017) 1607–1614, <http://dx.doi.org/10.1007/s10620-017-4539-5>.
- [12] D.F. Berg, A.M. Bahadursingh, D.L. Kaminski, W.E. Longo, Acute surgical emergencies in inflammatory bowel disease, Am. J. Surg. 184 (2002) 45–51.
- [13] L. Bollen, N. Vande Casteele, V. Ballet, G. Van Assche, M. Ferrante, S. Vermeire, A. Gils, Thromboembolism as an important complication of inflammatory bowel disease, Eur. J. Gastroenterol. Hepatol. 28 (2016) 1–7, <http://dx.doi.org/10.1097/MEG.0000000000000495>.
- [14] N.L. Zitomersky, M. Verhave, C.C. Trenor, Thrombosis and inflammatory bowel disease: a call for improved awareness and prevention, Inflamm. Bowel Dis. 17 (2011) 458–470, <http://dx.doi.org/10.1002/ibd.21334>.

**Open Access**

This article is published Open Access at [sciencedirect.com](https://www.sciencedirect.com). It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.