



Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Pylephlebitis and Crohn's disease: A rare case of septic shock

Stefano Scaringi^a, Francesco Giudici^{a,*}, Giacomo Gabbani^b, Daniela Zambonin^a, Marco Morelli^c, Rossella Carrà^c, Paolo Bechi^a^a Digestive Surgery Unit, Department of Surgery and Translational Medicine, Careggi University Hospital, Largo Brambilla 3, 50134 Florence, Italy^b Diagnostic and Operative Radiology, Department of Emergency, Careggi University Hospital, Italy^c Intensive Care Medicine, Ospedale del Mugello, Azienda Sanitaria Firenze, Italy

ARTICLE INFO

Article history:

Received 18 June 2017

Received in revised form 29 June 2017

Accepted 8 August 2017

Available online 10 August 2017

Keywords:

Crohn's disease

Pylephlebitis

Septic shock

Gastroenterology

Surgery

ABSTRACT

INTRODUCTION: Troncular pylephlebitis, defined as septic thrombophlebitis of the portal vein, is usually secondary to suppurative infection from the regions drained by the portal system. Therefore, pylephlebitis can occur from the portal vein main tributaries. The occurrence of mesenteric pylephlebitis in Crohn's disease is extremely rare.

PRESENTATION OF CASE: We describe a case of septic shock due to mesenteric pylephlebitis in a 47 years old male affected with Crohn's disease. The patient was admitted to the emergency department after he had been complained from 3 h of a *peri*-umbilical abdominal pain associated to fever and shivering quickly followed by a severe hypotension. His medical history included histologically confirmed ileal Crohn's disease diagnosed 4 years before and treated with mesalamine only. Computed tomography scan confirmed the mesenteric pylephlebitis diagnosis. After medical therapy with antibiotics and systemic nutrition, the patient was successfully operated to treat his ileal Crohn's disease.

DISCUSSION: In our case, the quick onset of a septic shock was not due to a peritonitis complicating a Crohn's disease, but to a rare condition not needing an urgent surgical resolution. This report shows that, even in Crohn's disease, once diagnosis is performed, antibiotic therapy associated to enteral and parenteral nutrition can lead to a complete clinical remission of mesenteric pylephlebitis, mandatory to perform an elective surgery.

CONCLUSION: This case highlights the importance of promptly considerate and treat mesenteric pylephlebitis in presence of a septic shock in a Crohn's disease patient who is not showing clinical signs of peritonitis.

© 2017 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

Pylephlebitis, defined as septic thrombophlebitis of the portal vein system, is usually secondary to a suppurative process developed in the region drained by the portal vein (troncular pylephlebitis) or by its main tributaries (mesenteric or splenic pylephlebitis) [1]. Pylephlebitis was diagnosed at autopsy and described for the first time by Waller in 1846 in a patient affected with appendicitis [2]. In the past its prognosis was extremely poor, but broad-spectrum antibiotics and surgical removal of the infective focus were able to decrease both its incidence and mortality.

Pylephlebitis was described as a Crohn's disease complication in 1946 by Taylor [3] and, to the best of our knowledge, only other 8 cases have been reported so far [2,4–8]. Furthermore, only in three of them the superior mesenteric vein was described to be primarily involved. We report a case of septic shock due to mesenteric pylephlebitis in a patient affected with recently diagnosed ileal Crohn's disease. This work has been reported in line with the SCARE criteria [9].

2. Presentation of case

A 47-year-old man was admitted to the emergency department because of a septic shock. He had been complained from 3 h of a *peri*-umbilical abdominal pain associated to fever and shivering. These symptoms were quickly followed by a severe hypotension. His medical history included histologically confirmed ileal Crohn's disease diagnosed 4 years before and treated with mesalamine only. He was 187 cm tall and 92 kg weight. Physical examination revealed a fixed *peri*-umbilical mass without abdominal guard-

* Corresponding author.

E-mail addresses: stefano.scaringi@unifi.it (S. Scaringi), francesco.giudici@unifi.it (F. Giudici), giacomogabbani@gmail.com (G. Gabbani), daniela.zambonin@gmail.com (D. Zambonin), marco.morelli@uslcentro.toscana.it (M. Morelli), rossella.carra@uslcentro.toscana.it (R. Carrà), paolo.bechi@unifi.it (P. Bechi).

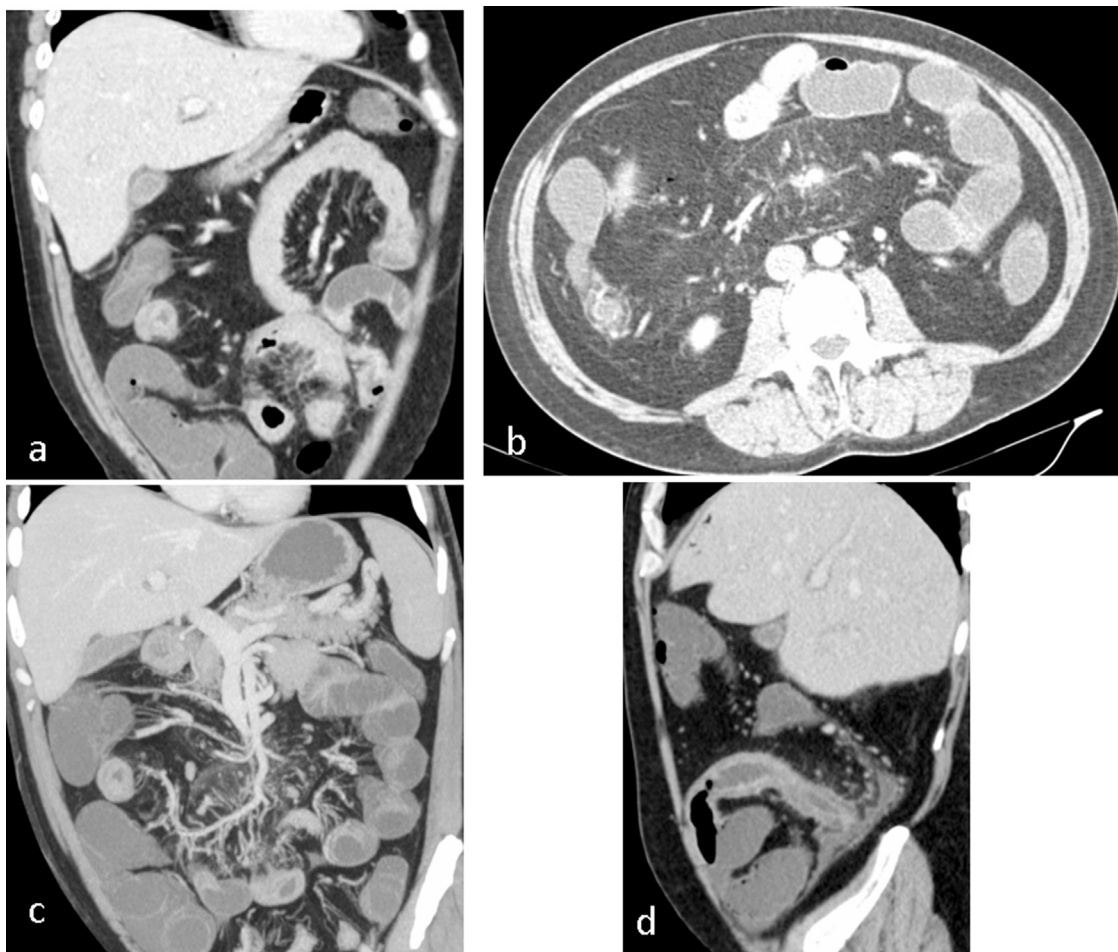


Fig. 1. Thickening and alteration of the intestinal wall (1a); small air bubbles were visible in the mesentery (1b). Fat stranding with obstruction of the inferior mesenteric vein (1c); peripheral linear collections of gas in the liver (1d).

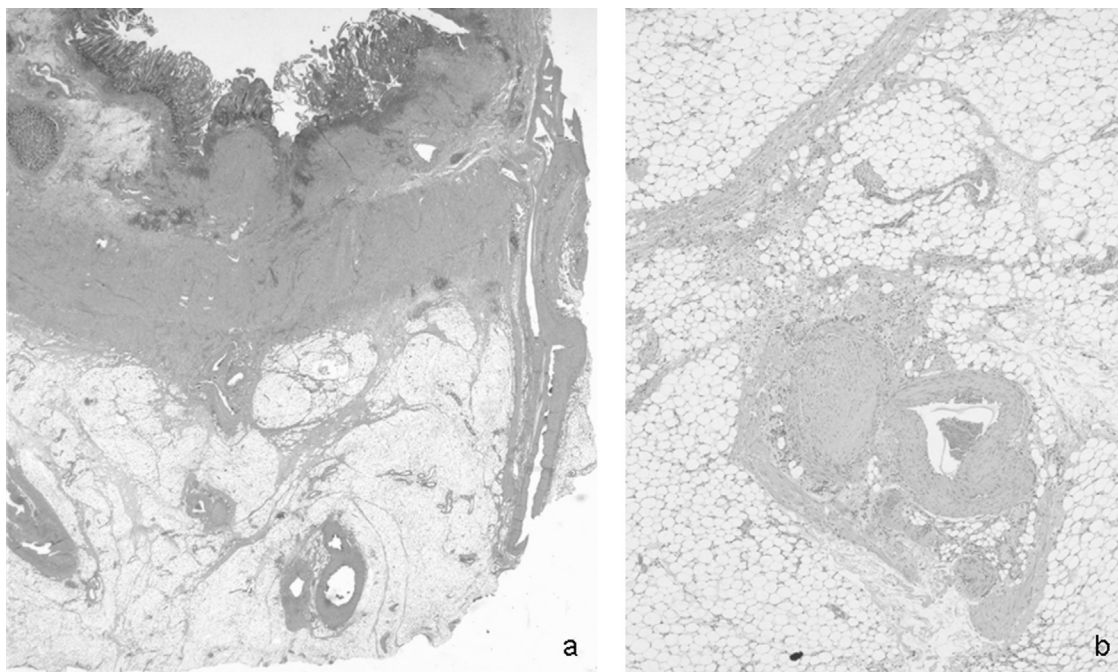


Fig. 2. Crohn's disease of the ileum with associated obliterative vasculopathy: haematoxylin and eosin stain x 20 HPF (a); haematoxylin and eosin stain x100 HPF (b).

ing. Blumberg sign was negative. Initial workup revealed severe leukopenia (300 white blood cells/mcL) and high procalcitonin levels (453 µg/L). Abdominal x-ray excluded an intestinal perforation. Abdominal computed tomography (CT) scan was performed (Fig. 1): it confirmed the presence of a Crohn's disease involving the terminal ileum for a length of about 70 cm. Along this small bowel tract the mesentery was extremely inflamed as in a phlegmonous condition causing the obstruction of the inferior mesenteric vein. Air bubbles were visible both in the mesentery and in the liver. These findings were suggestive for mesenteric pylephlebitis complicating ileal Crohn's disease. The patient was in a severe septic shock but without having clinical sign of abdominal perforation and peritonitis. Both leukopenia and high procalcitonin value suggested the presence of Gram-negative bacteremia. Patient was promptly supported by mechanical ventilation and vasopressors agents. A broad-spectrum antibiotic therapy (intravenous carbapenem: meropenem 1 gr per 3 times/day) was administered and the catabolic state corrected with both enteral and parenteral nutriments. Peripheral blood culture showed the presence of *Klebsiella oxytoca* and *Escherichia coli*. Antibiotics and fluids resuscitation brought the patient to a quick improvement of his general conditions and after 3 weeks in the intensive care unit the patient was discharged. He underwent elective surgery about two months later. At surgery the mesentery of the terminal ileum was extremely retract and increased in its thickness (more than 8 cm), with some diseased ileal loops tangentially attached to it. An ileo-colic resection (80 cm + 7 cm) was performed, followed by an ileo-colic side to side anastomosis. Post-operative course was uneventful and the patients discharged at day 10. Histology confirmed Crohn's disease of the terminal ileum associated to an obliterative vasculopathy (Fig. 2). Biological therapy was started and after 6 months follow-up the patient is in good general conditions and free from recurrence.

3. Discussion

Mesenteric pylephlebitis is an infective suppurative thrombosis of the mesenteric vein or its branches. It is usually secondary to the development of intra-abdominal infective foci typically complicating diverticular disease, appendicitis and necrotizing pancreatitis [2]. This critical clinical condition can arise in inflammatory bowel diseases too [2–8]. However, in Crohn's disease it is extremely rare. Furthermore, as showed by this report, the occurrence of such a complication seems not to be directly related to the severity of the Crohn's disease: our patient was affected by a mild form of Crohn's disease, recently diagnosed and treated with mesalazine only.

The patient experienced septic shock without clinical signs of peritonitis: we believe in Crohn's disease it is extremely important mesenteric pylephlebitis to be considered in presence of such clinical presentation, since nowadays, as showed by this report, through a resuscitation therapy associated to broad-spectrum antibiotics and enteral and parenteral nutrition, it is a curable condition even in presence of concomitant Crohn's disease. Two months after the patient had been discharged from the intensive care unit, we indicated surgery, performing the ileo-colic resection with ileo-colic side to side anastomosis. We believe the strict clinical evaluation by surgeons, gastroenterologists and anesthesiologists was extremely important allowing the patient to be electively operated in a "window" period characterized by a good global health status, starting from an optimal nutritional repletion, auspicious especially in patients affected with Crohn's disease, to obtain an uneventful postoperative outcome, as well as to minimize the risk of post-operative stoma [10]. Interestingly, histological analysis on the surgical specimen confirmed the presence of an obliterative vasculopathy, even in absence of the initial clinical symptoms.

4. Conclusion

Multidisciplinary approach is mandatory for the decision making process in presence of mesenteric pylephlebitis, to obtain both a correct diagnosis and a prompt treatment. In fact, although surgical approach is often necessary, timing for surgery is very important in order to operate the patient in an elective setting, which is mandatory to perform a bowel sparing surgery, minimizing the intra and post-operative complications.

Conflict of interest

None.

Funding

None.

Ethical approval

Not needed.

Consent

It was obtained from the involved patient.

Author contributions

All Authors contributed equally to conception and design, and/or acquisition of data, and/or analysis and interpretation of data for this work; all Authors participated in drafting the article or revising it critically for important intellectual content and gave final approval of the version to be submitted and any revised version. Acquisition of data: Scaringi, Giudici, Gabbani, Zambonin, Morelli, Carrà; Analysis and interpretation of data: Scaringi, Gabbani, Giudici, Bechi; Drafting of manuscript: Scaringi, Giudici, Zambonin, Bechi; Critical revision: Scaringi, Gabbani, Giudici, Morelli, Carrà.

Guarantor

Dott. Stefano Scaringi.

Acknowledgment

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

References

- [1] J.J. Drabick, F.J. Landry, Suppurative pylephlebitis, *S. Med. J.* 84 (1991) 1396–1398.
- [2] J.W. Baddley, D. Singh, P. Correa, N.J. Persich, Crohn's disease presenting as septic thrombophlebitis of the portal vein (pylephlebitis): case report and review of the literature, *Am. J. Gastroenterol.* 94 (1999) 847–849.
- [3] F.W. Taylor, Regional enteritis complicated by pylephlebitis and multiple liver abscesses, *Am. J. Med.* 7 (1949) 838–840.
- [4] A.R. Shin, C.K. Lee, H.J. Kim, J.J. Shim, J.Y. Jang, S.H. Dong, B.H. Kim, Y.W. Chang, Septic pylephlebitis as a rare complication of Crohn's disease, *Korean J. Gastroenterol.* 61 (2013) 219–224.
- [5] M. Aguas, G. Bastida, P. Nos, B. Beltrán, J.L. Grueso, J. Grueso, Septic thrombophlebitis of the superior mesenteric vein and multiple liver abscesses in a patient with Crohn's disease at onset, *BMC Gastroenterol.* 7 (2007) 22.
- [6] J.Y. Tung, J.L. Johnson, C.A. Liacouras, Portal-mesenteric pylephlebitis with hepatic abscesses in a patient with Crohn's disease treated successfully with anticoagulation and antibiotics, *J. Pediatr. Gastroenterol. Nutr.* 23 (1996) 474–478.

- [7] B. Lerman, J.H. Garlock, H.D. Janowitz, Suppurative pylephlebitis with multiple liver abscesses complicating regional ileitis: review of literature-1940–1960, *Ann. Surg.* 155 (1962) 441–448.
- [8] S. Kluge, K.E. Hahn, C.H. Lund, A. Gocht, G. Kreymann, Pylephlebitis with air in the portal vein system. An unusual focus in a patient with sepsis, *Dtsch. Med. Wochenschr.* 128 (2003) 1391–1394.
- [9] R.A. Agha, A.J. Fowler, A. Saeta, I. Barai, S. Rajmohan, D.P. Orgill, SCARE Group, The SCARE statement: consensus-based surgical case report guidelines, *Int. J. Surg.* 34 (2016) 180–186.
- [10] R.R. Cima, B.G. Wolff, Reoperative Crohn's surgery: tricks of the trade, *Clin. Colon Rectal Surg.* 20 (2007) 336–343.

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.