

HEMOPERITONEUM FOLLOWING MILD BLUNT ABDOMINAL TRAUMA: FIRST PRESENTATION OF CROHN'S DISEASE

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ABSTRACT

Introduction: Inflammatory bowel diseases are heterogeneous in their presentation. Crohn's disease (CD) has been described as an unusual cause of massive lower gastrointestinal bleeding. We present a patient with CD whose first presentation was hemoperitoneum following mild blunt abdominal trauma

Case presentation: A 54-year-old woman came to the emergency room with a blunt abdominal trauma. The patient underwent an abdominal CT scan with i.v. contrast medium injection, which showed hemoperitoneum with stenotic strictures of the small bowel and active massive bleeding from ileocolic artery. The rapid anemia onset and the CT report induced us to perform blood transfusion and an emergency exploratory laparotomy. Histological examination of the resected terminal ileum revealed a Crohn's disease.

Discussion: Crohn's disease has been associated with hemoperitoneum, but to date there is no exact protocol regarding treatment of massive hemorrhagic Crohn's disease, which is rare. A conservative approach with medical therapy has been suggested for initial treatment. However, if medical treatment fails or bleeding continues even with intervention, bowel resection through surgical therapy should be performed.

Key words: Crohn's disease, hemoperitoneum, emergency surgery, blunt abdominal trauma, CT scan.

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Introduction

Inflammatory bowel diseases (IBD) are idiopathic chronic diseases of the gastrointestinal tract well known to be associated with both genetic and environmental risk factors and with larger diffusion in Western countries. Although the exact aetiology of IBD has still not been exactly identified, it is believed that the pathogenesis of IBD includes immune deregulation secondary to environmental factors in genetically susceptible individuals. Modern studies report also a role of particular protein, like chaperonins, in the activation of the immune system leading to inflammation⁽¹⁾. Some epidemiological studies have already identified the increasing global incidence and prevalence of IBD in developing countries as these countries become

more developed and "Westernised", highlighting the significance of environmental factors on influencing IBD development globally. Currently, only smoking is an established risk factor for Crohn's disease (CD). There seems to be a dose-response relationship between smoking and IBD and childhood exposure to tobacco smoke is associated with a higher risk of developing CD. Oral contraceptive pill increases the risk of IBD too. Regarding diet, the literature is inconsistent and no clear dietary risk factors for IBD can be determined. IBD are heterogeneous in their presentation. The most frequent symptoms are chronic diarrhea with blood and/or mucus, recurrent abdominal pains or discomfort, melena, weight loss, and/or perianal fistula or abscess. CD is diagnosed if we find at endoscopy skip lesions, a cobblestone appearance, mucosal

ulceration and/or aphthous lesions. Extraintestinal manifestations include musculoskeletal (sacroilitis and peripheral arthritis), mucocutaneous (erythema nodosum, pyoderma gangrenosum, aphthous stomatitis), hepatic (primary sclerosing cholangitis), ophthalmic (episcleritis, uveitis) and urinary tract involvements. CD has been described as an unusual cause of massive lower gastrointestinal bleeding occurring only in 0.9 - 6% of patients^(2,3,4). CD significantly alters endothelial and vascular function and this contributes to uncontrolled vascular-dependent intestinal damage^(5,6). This is a hallmark of active gut disease and is closely related to disease severity.

We present a patient with CD whose first presentation was hemoperitoneum following mild blunt abdominal trauma.

Case presentation

A 54-year-old woman came to the emergency room with a blunt abdominal trauma. On arrival she complained of worsening abdominal pain. On physical examination she was slightly pale: heart rate 88/min and blood pressure of 100/50 mmHg. Abdominal examination revealed tympanic bowel sounds in the lower abdominal quadrants without signs of peritonitis. Routine blood test showed Hb 12.6 g/dl; RBC 4.48 x 10⁶ with increasing of markers of inflammation (WBC 15.23 x 10³; Neutrophil count 90.2%; C-reactive Protein 87 mg/l). She had a medical history of appendectomy, removal of ovarian cyst and temporo-parietal meningioma excision.

The patient underwent an abdominal CT scan with i.v. contrast medium injection, which showed hemoperitoneum: in particular, blood effusion assumed a triangular shape as it insinuated between small bowel loops. Moreover, on CT it was notable the presence of stenotic strictures of the small bowel, with increased mucosal enhancement, mesenteric inflammatory stranding and mesenteric adenopathy. These signs were typical features of CD patients that we could analyze in clinical routine (e.g. use of the computed tomography enterography in evaluation of CD activity)⁽⁷⁾ (Figure 1). In this clinical case the CT showed also, in portal and late phases, tortuous mesenteric arteries, with suspect extravasation from the ileocolic artery. On the basis of these findings, we suggested a diagnosis of hemoperitoneum due to the rupture of a branch of ileocolic artery in a patient with CD (Figure 2).

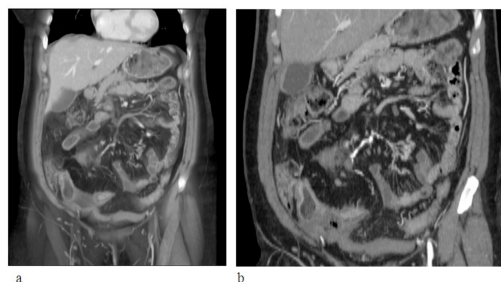


Fig. 1: a) 3D VR MDCT reconstruction shows the small wall thickening and strictures of the terminal ileum and the mesenteric bleeding. b) coronal reconstruction shows the bleeding from a branch of mesenteric ileocolic vein.

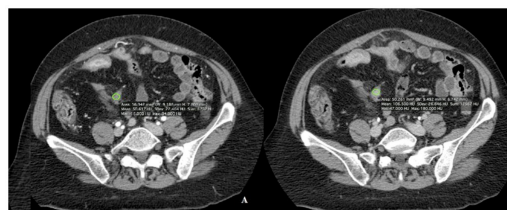


Fig. 2: MDCT axial scan in arterial (a) and venous (b) phases shows the progressive mesenteric bleeding from a branch of mesenteric ileocolic vein.

So, the patient was admitted to the Surgery Department. Heart rate and blood pressure were normal but the control blood test showed us a significant decline in hemoglobin (Hb 6.8 g/dl vs 12.6 g/dl; WBC 12 x 10³; Neutrophil count 85.8%; C-reactive Protein 95 mg/l). In other patients with hemoperitoneum or abdominal trauma we used successfully laparoscopic approach in emergency setting⁽⁸⁾, but in this case the rapid anemia, the development of severe hypotension and the CT report (hemoperitoneum due to the rupture of a branch of ileocolic artery) induced us to perform blood transfusion and an emergency exploratory laparotomy⁽⁹⁾ without any else study for cardiac or anesthesiologic risk stratification⁽¹⁰⁾.

At laparotomy it was clear the presence of blood effusion with clots and full-thickness mesenteric tear with active bleeding from ileocolic vessels. Moreover, it was found that a considerable length (30 cm proximal to the ileocecal valve) of the distal small bowel loops was thickened and showed interloop adhesions with edematous mesentery, as it appears in IBDs. Considering both the hemoperitoneum and the findings suggestive for IBD, the patient underwent a right hemicolectomy extended to the distal ileum (40 cm) and ileo-transverse mechanical termino-lateral anastomosis. The

specimen was then analyzed by an experienced pathologist. Macroscopic evaluation showed that ileal loops were adhering firmly to each other for a length of 32 cm; on the surface there was a focal hemorrhagic area. The mesentery was thickened and its surface was irregular and brownish. On opening the specimen, there was narrowing of the downstream lumen for a length of 12 cm and bowel-wall thickening characteristic of CD. The mucosa was diffusely eroded showing the characteristic cobblestone appearance up to the ileocecal valve, for a length of 22 cm.

Histological examination of the resected terminal ileum revealed areas of denuded mucosa and inflammatory cell infiltration and lymphoid aggregates in the lamina propria and, sometimes, in the submucosa. The mesenteric lymph nodes showed reactive hyperplasia (Figure 3).

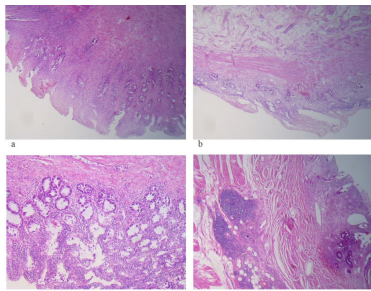


Fig. 3: a-b) EE 4x, areas of disepithelization of the colonic mucosa; c-d) EE 10x, inflammation in the lamina propria extending focally into the submucosa with infiltrate of lymphocytes, neutrophil and eosinophil granulocyte.

Discussion

Acute gastrointestinal hemorrhage is a rare presentation of CD^(2, 3, 4). CD alters endothelial and vascular function and this contributes to uncontrolled vascular-dependent intestinal damage^(5, 6, 11). As recently proved by Kohoutova et al.⁽⁶⁾ acquired hypercoagulable abnormalities in IBD patients are frequent. Funayama et al.⁽¹²⁾ reported significant atrophy in the media of the submucosal peripheral arteries in patients suffering from ileal CD. As stated by Cromer et al.⁽⁵⁾, many inflammatory growth factors, cytokines and chemokines mediate angiogenesis in IBD: the newly formed endothelium and the inflamed vessels differs from the normal ones in the production of and response to inflammatory cytokines, growth factors, and adhesion molecules, altering coagulant capacity, barrier function and blood cell recruitment in injury. Modern studies report also a role of particular protein, like chaper-

onins, in the activation of the immune system. This is a hallmark of active gut disease and is closely related to disease severity. Since 1970 CD has been associated to hemoperitoneum⁽¹³⁾, but to date there is no exact protocol regarding treatment of massive hemorrhagic CD, which is rare occurrence⁽¹⁴⁾.

A conservative approach with medical therapy has been suggested for initial treatment⁽¹⁵⁾. However, if medical treatment fails or bleeding continues even with intervention, bowel resection through surgical therapy should be performed⁽¹⁶⁾.

Our patient had never complained symptoms of IBD and this was her first presentation, following blunt abdominal trauma. CT scan prompted a provisional diagnosis of CD and blood examinations with rapid anemia due to massive hemorrhage induced us to the decision of emergency laparotomy.

The presence of CD in patients presenting like ours, with obvious trauma and absence of other symptoms, is unlikely to be suspected and diagnosis could only be made after CT scan, laparotomy and histology^(17, 18). Moreover, Brownstein⁽¹⁹⁾ reported the case of an acute small bowel obstruction occurring five months after blunt abdominal trauma, in which the histopathology of the affected segment of ileum was found to closely resemble CD. Burak et al.⁽²⁰⁾ reported also a case of suspected CD at laparoscopy, not confirmed at enteroscopy, in a woman with recurrent attacks of abdominal pain following minor abdominal trauma, and whose final diagnosis was C1 inhibitor deficiency.

These findings highlight the difficulty in the diagnosis of CD in case of abdominal trauma.

In our patient, the presence of an inflamed edematous mesentery and tortuous mesenteric arteries as complications of undiagnosed and untreated CD made her prone to hemorrhage.

References

- 1) Rodolico V, Tomasello G, Zerilli M, Martorana A, Pitruzzella A, Gammazza AM, David S, Zummo G, Damiani P, Accomando S, Conway de Macario E, Macario AJ, Cappello F: *Hsp60 and Hsp10 increase in colon mucosa of Crohn's disease and ulcerative colitis*. Cell Stress Chaperones. 2010 Nov; 15(6): 877-84.
- 2) Cirocco W.C., Reilly J.C., Rusin L.C. *Life-threatening haemorrhage and exsanguination from Crohn's disease. Report of four cases*. Dis Colon Rectum. 1995; 38: 85-95.
- 3) Driver C.P., Anderson D.N., Keenan R.A. *Massive intestinal bleeding in association with Crohn's disease*. J R Coll Surg Edinb. 1996; 41: 152-154.

- 4) Majeed MU, Jameel M, Williams DT *Massive per rectal bleeding following blunt abdominal trauma: First presentation of Crohn's disease*. Int J Surg Case Rep. 2011; 2(7): 230-231.
- 5) Cromer WE, Mathis JM, Granger DN, Chaitanya GV, Alexander JS. *Role of the endothelium in inflammatory bowel diseases*. World J Gastroenterol. 2011 Feb 7; 17(5): 578-93.
- 6) Kohoutova D, Pecka M, Cihak M, Cyrany J, Maly J, Bures J. *Prevalence of hypercoagulable disorders in inflammatory bowel disease*. Scand J Gastroenterol. 2013 Dec 16. [Epub ahead of print]
- 7) Lo Re G, Cappello M, Tudisca C, Galia M, Randazzo C, Craxì A, Cammà C, Giovagnoni A, Midiri M. *CT enterography as a powerful tool for the evaluation of inflammatory activity in Crohn's disease: relationship of CT findings with CDAI and acute-phase reactants*. Radiol Med. 2014 Jan. [Epub ahead of print].
- 8) Agrusa A, Romano G, De Vita G, Frazzetta G, Chianetta D, Di Buono G, Gulotta G. *Adrenal gunshot wound: Laparoscopic approach. Report of a case*. Int J Surg Case Rep. 2014; 5(2): 70-2.
- 9) Perino A, Proto E, Calagna G, Granese R, Agrusa A, Guarneri F, Cucinella G: *Spontaneous rupture of splenic artery aneurysm in pregnancy: is splenectomy always necessary?* Acta Obstet Gynecol Scand. 2012 Nov; 91(11): 1349-50.
- 10) Novo G, Corrado E, Tortorici E, Novo A, Agrusa A, Saladino V, Marturana I, Lentini R, Ferrandes M, Visconti C, Massenti F, D'Arienzo M, Vitale F, Gulotta G, Novo S. *Cardiac risk stratification in elective non-cardiac surgery: role of NT-proBNP*. Int Angiol. 2011 Jun; 30(3): 242-6.
- 11) Leonetti D, Reimund JM, Tesse A, Viennot S, Martinez MC, Bretagne AL, Andriantsitohaina R. *Circulating microparticles from Crohn's disease patients cause endothelial and vascular dysfunctions*. PLoS One. 2013 Sep 3; 8(9): e73088. doi: 10.1371/journal.pone.0073088.
- 12) Funayama Y, Sasaki I, Naito H, Fukushima K, Matsuno S, Masuda T. *Remodeling of Vascular Wall in Crohn's Disease*. Dig Dis Sci. 1999 Nov; 44(11): 2319-23.
- 13) McCullough FS, Ruppert RD, Greenberger NJ. *Recurrent Hemoperitoneum. An Unusual Complication of Regional Enteritis* Arch Intern Med. 1970; 126(3): 514-6.
- 14) Iiritano E, Grassia R, Staiano T, Buffoli F. *Life-threatening jejunal hemorrhage as first presentation of Crohn's disease*. Inflamm Bowel Dis. 2010; 16: 1277-1278.
- 15) Kazama Y, Watanabe T, Akahane M, Yoshioka N, Ohtomo K, Nagawa H. *Crohn's disease with life-threatening hemorrhage from terminal ileum: successful control by superselective arterial embolization*. J Gastroenterol. 2005; 40: 1155-1157.
- 16) Agrusa A, Romano G, Di Buono G, Dafnomili A, Gulotta G. *Laparoscopic approach in abdominal emergencies: a 5-year experience at a single center*. G Chir. 2012; 33(11-12): 400-3.
- 17) Lo Re G, Galia M, Bartolotta TV, Runza G, Taibbi A, Lagalla R, De Maria M, Midiri M. *Forty-slice MDCT enteroclysis: evaluation after oral administration of isotonic solution in Crohn's disease*. Radiol Med. 2007; 112(6): 787-97.
- 18) Romano G, Agrusa A, Frazzetta G, De Vita G, Chianetta D, Di Buono G, Amato G, Gulotta G: *Laparoscopic drainage of liver abscess: case report and literature review*. G Chir. 2013 May-Jun; 34(5-6): 180-2.
- 19) Brownstein EG. *Blunt abdominal trauma simulating Crohn's disease of the terminal ileum*. Aust N Z J Surg. 1984; 54(3): 287-9.
- 20) Burak KW, May GR. *C1 inhibitor deficiency and angioedema of the small intestine masquerading as Crohn's disease*. Can J Gastroenterol. 2000 Apr; 14(4): 349-451.

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