



Case report

Lower extremity necrotizing fasciitis: A unique initial presentation of Crohn's disease[☆]Anna Weiss^{a,*}, Bryan Sandler^b^a University of California, San Diego, 200 West Arbor Drive, MC 8402, San Diego, CA 92103, USA^b Center for the Future of Surgery, UCSD, 9500 Gilman Drive MC 0740, Medical Education and Telemedicine Building, La Jolla, CA 92093, USA

HIGHLIGHTS

- A patient with necrotizing fasciitis caused by an intraabdominal process.
- Fistulizing Crohn's disease first presented as necrotizing fasciitis.
- Hip disarticulation in a crohn's patient.

ARTICLE INFO

Article history:

Received 10 April 2015

Accepted 18 May 2015

Keywords:

Crohn's disease

Necrotizing fasciitis

ABSTRACT

Crohn's disease is a disease of the bowel, typically presenting with diarrhea, weight loss, and abdominal pain. Complications such as abscesses, fistulas, and strictures may require surgical intervention. We would like to report a patient with Crohn's disease who presented for the first time with left lower extremity necrotizing fasciitis. There are very few reports of necrotizing fasciitis in Crohn's disease as the initial presentation.

© 2015 Published by Elsevier Ltd on behalf of IJS Publishing Group Limited. 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 primarily a disease of the bowel, which typically presents with diarrhea, weight loss, and abdominal pain. It most commonly affects the distal ileum and colon. It is characterized by extensive fibrosis, strictures, and fistulas [1]. Complications such as abscesses, fistulas, and strictures may require surgical intervention. We would like to report a patient with Crohn's disease, who primarily presented with left lower extremity necrotizing fasciitis. There are very few reports of necrotizing fasciitis in Crohn's disease [2], none in which fasciitis was the primary presenting symptom.

2. Case report

A 62 year old man presented to the Emergency Department with three weeks of left thigh edema, new onset erythema and excruciating pain to light touch. He had no known past medical history, was taking no medications, and was a heavy smoker. The patient was cachectic on general examination. He was tachycardic with

elevated white cell count and erythrocyte sedimentation rate. A computed tomography (CT) scan was obtained which revealed extensive subcutaneous gas and fluid from the anterior superior iliac spine to the iliopsoas, and circumferentially along all fascial planes. Additionally, a small fluid collection was seen along the left psoas muscle bed; no other fluid collections were noted radiographically. General Surgery was consulted and the patient was brought emergently to the operating room. The anterior and posterior compartments contained a significant amount of purulent, foul-smelling fluid, and the degree of necrosis necessitated hip disarticulation (Fig. 1).

The patient was managed with broad spectrum antibiotics, including vancomycin, piperacillin-tazobactam, metronidazole, and clindamycin. Cultures were poly-organismal, growing *Escherichia coli*, *Viridans streptococci*, *Fusobacterium* species, *Bacteroides fragilis*, *Prevotella* species, and *Clostridium* species (not *perfringens*) from the thigh. The pelvis grew the same bacteria with the addition of *Peptostreptococcus* species. Antibiotics were tailored to piperacillin-tazobactam alone when he clinically improved. A bowel source, possible perforation was always suspected; however the patient never complained of abdominal pain and consistently had a benign exam. To investigate the source of the poly-organismal infection, multiple CT scans of the abdomen were obtained which initially revealed only inflamed terminal ileum,

[☆] Research registered at www.researchregistry.com, UIN researchregistry114.

* Corresponding author.

E-mail addresses: a3weiss@ucsd.edu (A. Weiss), bsandler@ucsd.edu (B. Sandler).



Fig. 1. Surgical specimen after hip disarticulation. The anterior and posterior compartments contained a significant amount of purulent, foul-smelling fluid. Extensive necrosis was evident up to the level of the iliopsoas.

providing no definite explanation.

On second look, minimal debridement of the wound was necessary. A vacuum dressing was applied, several days later this expressed feculent material. A barium enema and small bowel follow-through revealed thickened loops of terminal ileum, and multiple ileo-sigmoid fistulas – a source identified for the necrotizing infection of the lower extremity. The fistula involving the terminal ileum and sigmoid colon was resected, and a diverting ileostomy created. Pathology of the surgical specimen revealed Crohn's disease. The patient had an uneventful post-operative course.

The patient's Crohn's disease later presented with perianal fistula disease. The patient spent two years in physical therapy, being treated medically for his Crohn's disease, working on smoking cessation, and improving his nutritional status. The patient's top priority was walking again, however the ileostomy made fitting a lower extremity prosthesis, which required waist fitting, impossible. Once the patient successfully quit smoking and had minimal perianal symptoms, his ileostomy was reversed. He had no peri-operative complications, and recovered well.

3. Discussion

Crohn's disease is a disease of unknown etiology. Many theories exist including impaired intestinal epithelial barrier function, disturbed immunological response, and an as-of-yet unidentified infection [1]. Mainstay of treatment is medical therapy, with surgery for refractory symptoms and complications. Progressive or uncontrolled symptoms despite maximal medical treatment, intolerable medication side effects, or poor patient adherence are

considered failures of medical therapy. Fistulas with associated abscesses or strictures require surgery [3]. Despite the more aggressive and earlier use of immunosuppressants, the need for surgery has not decreased. Nearly 70–90 percent of Crohn's patients will need surgery despite medical advances [4].

Fistula disease occurs in up to 50% of patients with Crohn's [5,6]. Crohn's fistula disease presents predominantly as perianal disease (about 50%), and less often as entero-enteric disease (about 25%) [5]. Crohn's fistula differ in the inflammatory profile as compared to non-Crohn's fistula – both have macrophage infiltration but Crohn's disease has more abundant T and B cell infiltration [5]. Healing rates for are very low for these fistulae [7,8]. In one study [7], median healing time for a fistula was 2.5 years, 68% healed eventually, 18% only partially healed and 14% did not heal. A combined medical and surgical approach to the Crohn's fistula is best. 90% of all fistulae will require surgery [6]. Diagnosis relies on physical exam, exam under anesthesia when necessary, and rarely an endorectal ultrasound may be of use. Medical treatment includes antibiotics, immunomodulators and biologic therapy – specifically anti-TNF- α drugs. Surgical treatments start with simple incision and drainage, and progress to seton placement, fistulotomy, fistula plugs, endorectal advancement flaps, ligation of the intersphincteric tract, to most severe diversion or proctectomy [6].

We reason that this patient's necrotizing fasciitis was due to bacterial translocation from his ileo-sigmoid fistulating disease burden. The path of disease may have been along his psoas muscle, or retroperitoneum and femoral canal. It is unclear why his intra-abdominal disease remained otherwise asymptomatic, until stool began to accumulate in the wound vac. He had poor nutritional status on presentation, and was also a heavy smoker. Smoking is a risk factor for Crohn's patients to undergo major abdominal surgery, as well as a general predictor of worse outcome [9].

In conclusion, this patient had left lower extremity cellulitis and pain, which proved to be necrotizing fasciitis; however, had no abdominal complaints before or during his hospital stay. Given the lack of abdominal signs or symptoms and relatively benign CT scans, this patient presents a unique challenge to diagnose Crohn's disease earlier. This is the only report in which Crohn's disease was newly diagnosed after treatment of lower extremity necrotizing fasciitis.

Conflicts of interest

We have no conflicts of interest to report for this manuscript. Unrelated to this topic, but to guarantee full disclosure, Dr. Sandler is a consultant for ValenTX, Inc, and a speaker for Ethicon and Gore.

References

- [1] C. Leoward, G. Heuschen, P. Kienle, U. Heuschen, J. Schmidt, Surgical treatment of severe inflammatory bowel diseases, *Dig. Dis.* 21 (1) (2003) 54–62.
- [2] J.M. Kiely, K.J. Brasel, S.J. Graewin, D.M. Nowak, R. Nirula, Crohn's disease presenting as necrotizing fasciitis: a case report, *Am. Surg.* 71 (10) (2005 Oct) 870–871.
- [3] J.M. Hwang, M.G. Varma, Surgery for inflammatory bowel disease, *World J. Gastroenterol.* 14 (17) (2008 May 7) 2678–2690.
- [4] R. Caprilli, E. Angelucci, V. Clemente, Recent advances in the management of crohn's disease, *Dig. Liver Dis.* 40 (9) (2008 Sep) 709–716.
- [5] M. Scharl, G. Rogler, Pathophysiology of fistula formation in Crohn's disease, *World J. Gastrointest. Pathophysiol.* 5 (3) (2014 Aug 15) 205–212.
- [6] C.B. Geltzeiler, N. Wieghard, V.L. Tsikitis, Recent developments in the surgical management of perianal fistula for Crohn's disease, *Ann. Gastroenterol.* 27 (4) (2014) 320–330.
- [7] S.J. Bell, A.B. Williams, P. Wiesel, K. Wilkinson, R.C.G. Cohen, M.A. Kamm, The clinical course of fistulating Crohn's disease, *Aliment. Pharmacol. Ther.* 17 (9) (2003) 1145–1151.
- [8] S.A. Hyder, S.P. Travis, D.P. Jewell, N.J. McC Mortenson, B.D. George, Fistulating anal Crohn's disease: results of combined surgical and infliximab treatment, *Dis. Colon Rectum* 49 (12) (2006 Dec) 1837–1841.
- [9] C. Zallot, L. Peyrin-Biroulet, Clinical risk factors for complicated disease: how reliable are they? *Dig. Dis.* 30 (Suppl 3) (2012) 67–72.