Enterohepatic fistula in a Crohn's disease patient: A case report

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ABSTRACT

INTRODUCTION: Fistulous tracts are a hallmark of Crohn's Disease. However, solid organ to intestinal fistulas are rare with previously few case reports of colosplenic fistulas and one case report of an enterohepatic fistula.

PRESENTATION OF CASE: We review the available literature and present the first case report of an enterohepatic fistula in a female with Crohn's Disease to be treated operatively. The patient did well postoperatively with complete resolution of her fistula.

DISCUSSION: Crohn's Disease is an inflammatory bowel disease that can present with fistulas. However, a fistula between the liver and bowel is exceedingly rare with only one previous case report. This is the first report of an enterohepatic fistula that has been managed successfully with an operation.

CONCLUSION: Not all enterenteric fistulas are apparent preoperatively. When discovered, laparoscopic enterohepatic fistula takedown is feasible for this rare disease process manifestation.

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1. Introduction

Crohn's disease is an inflammatory bowel disease that is well known to cause fistulas with up to 35% of patients being affected [1]. In their cohort, the authors found that there were 88 fistulas in 59 patients. Thirty-nine (66%) of the 59 patients experienced one, 13 (22%) had two and 7 (12%) had three or more fistulizing episodes. Of the 88 fistulas, 48 (55%) were perianal, 21 (44%) were enterenteric, eight (9%) were rectovaginal, five (6%) were enterocutaneous, three (3%) were enterovesicular, and three (3%) were enterointra-abdominal fistulas.

Fistulas between a solid organ and intestine are rare, however, including case reports of colosplenic fistulas [2–4]. In Diana and George [3], the authors report a case of a 34 year old male with Crohn's who was found to have a fistula from his colon to his spleen. He was treated with a splenectomy and subtotal colectomy. There is one reported case in the Dutch literature of an enterohepatic fistula, which was not managed operatively. In their report, Rickes et al. describe a patient who developed a liver abscess from an enterohepatic fistula, similar to our case report. For their patient, the authors were not able to drain the abscess as it was not completely liquefied, but they gave the patient antibiotics and infliximab until the abscess resolved [5].

2. Presentation of case

A 32-year-old pregnant Caucasian female with a long-standing history of Crohn's Disease was seen as an inpatient consult at our academic, tertiary care, referral center for abdominal pain and fevers while in her third trimester of pregnancy. Workup included a CT-scan, which revealed severe terminal ileitis adherent to the right lobe of the liver along with a 3.9 cm × 3.5 cm hepatic abscess in the right lobe (Figs. 1 and 2). She underwent conservative management with antibiotics and Vascular Interventional Radiology drained the abscess percutaneously, as was standard of care. Unfortunately, the patient had a recurrent abscess concerning for a fistula between the abscess and small bowel. She then underwent CT enterography to rule out an enterohepatic fistula, which was negative for any fistulous tract.

She was counseled about her surgical options, which included both the laparoscopic and open approaches after failing conservative management, which necessitated surgery. She underwent an extensive laparoscopic lysis of adhesions, marsupialization of the liver abscess, and ileocecal resection a month later by an experienced laparoscopic colorectal surgeon. The inflamed, segment of terminal ileum was densely adhered to the liver. There were essentially no tissue planes and the terminal ileum was significantly adhered to the liver, as were the cecum and appendix. These were meticulously dissected off the visceral surface of the liver. When we dissected the diseased intestine off the liver, we found a purulent material coming from a small fistulous opening in the liver. We enlarged the fistulous opening and encountered a 5 cm × 6 cm abscess cavity filled with pus and enteric contents. This was ade-

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3. Discussion

Enterohepatic fistulas are exceedingly rare, especially when focusing on those due to Crohn’s disease. This patient is the first report in the English literature where operative intervention has been utilized to successfully treat the fistula. In our case, we were able to use a percutaneous drain to drain the abscess preoperatively, but this failed to completely treat it. While there are no treatment guidelines or trials comparing conservative versus operative management, case reports have demonstrated successful treatment with conservative measures. One important learning point from this case is that not all enteric fistulas are readily visible on imaging preoperatively and surgeons must consider this as a possibility for recurrent abscesses, especially in patients with inflammatory bowel disease. As in our case, some fistulas are found intraoperatively and require an understanding of how to adjust operative plans to address the fistula. Here, we have provided a conservative and operative treatment framework for future intra-abdominal enterohepatic fistula cases, which was not previously available in the literature.

4. Conclusion

An enterohepatic fistula with hepatic abscess is a very unusual manifestation of Crohn’s Disease, but here we present the first English case report. In this case, the patient required the operating room in order to resect the fistula as it failed conservative management. If a patient with an enterohepatic fistula requires operative intervention, we have shown that a laparoscopic approach can be successful with a good outcome.

Conflict of interest statement

The authors have no disclosures or conflicts of interest.

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Ethical approval

The Albany Medical Center Committee on Research Involving Human Subjects Institutional Review Board (IRB) has approved this case report. The report number is 4716.

Consent

The publication of this case report has been approved by the Albany Medical College Institutional Review Board.

The personal information of the patient in this case report has been removed so that it is not identifiable. This does not alter the scientific meaning of the reported case.

Authors contribution

Justin T. Van Backer: Contributed to drafting and gave final approval of the version to be published, in addition to contributions related to the retrieval of the information from the medical record. He is agreeable to be accountable for all aspects of the work, including the accuracy and integrity of the information and questions that may arise.

Edward C. Lee: Contributed to the drafting and editing, as well as gave final approval of the version to be published. He performed...
the initial evaluation, assessment and care of the patient. He is agreeable to be accountable for all aspects of the work in ensuring that questions that arise are investigated and the information is accurate.

Guarantors

Edward Lee and Justin Van Backer are Guarantors for this case report.

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