Case report

Colosplenic fistula presentation in the context of undiagnosed colon cancer: Case report and review of literature

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ABSTRACT

Introduction and importance: Enteric fistulas commonly arise from inflammatory, infectious, or neoplastic processes. Colosplenic fistulas are rare with only several reported worldwide. Case presentation: Herein, we present a case of colosplenic fistula in a 39-year-old gentleman with past history of rectal cancer previously in remission. He was admitted with severe abdominal pain and hemodynamic instability due to septic shock. The erect chest x-ray revealed pneumoperitoneum under the diaphragm. Clinical discussion: Laparotomy was performed and ileum perforation was managed by resection and anastomosis. The post-op recovery was complicated by a febrile episode. To locate the source of infection a contrasted abdominal computed tomography was ordered, confirming the presence of a splenic abscess, suggestive of colosplenic fistula. We proceeded with laparotomy for drainage of the abscess, with splenectomy and splenic flexure resections. He was discharged 40 days post-op at a pre-morbid state. Conclusion: Due to the high prevalence of colon cancer worldwide, novel complications such as the one reported here, are important to be reflected on. We hope this case can exemplify the significance of higher index of suspicion in at risk patient groups by the surgical teams and appropriate training on acute management of this rare complication.

1. Introduction

Colosplenic fistula is a tract formed between the inflamed colon and the spleen, it is a rare reason of splenic abscess formation. The diagnosis of fistula and concurrent abscess is difficult, and a CT scan and laparoscopic exploration may be required [1]. Determining the underlying causes of this rare condition can guide the treating team on the best treatment approach, with the surgical intervention remaining the mainstay treatment [2]. The choice for the colosplenic fistula management due to splenic flexure adenocarcinoma, is en-block resection [3].

Herein, we discuss a rare case of colosplenic fistula in a patient with previous history of colon cancer and discuss nuances of diagnosis and treatment, intending to educate and raise the degree of suspicion for this rare complication in a particular patient demographic. This case report is in line with SCARE Criteria [4]. As far as we are aware, there has been very few similar cases in the literature, which makes this case novel and a useful educational tool.

2. Case presentation

A previously well 39-year-old documentary director living independently at home with his wife, presented to the emergency department complaining of one-week long progressive epigastric pain with associated nausea, vomiting and constipation. His past medical history was significant for rectal adenocarcinoma which was previously managed by abdomino-perineal resection (APR), end colostomy surgery and adjuvant chemoradiation therapy, 13 years ago. He had no other medical conditions and at the time of presentation was not taking any medications. He was a non-smoker and non-drinker.

On arrival he was clinically in septic shock. He had a fever his temperature being 39 °C [1–13], was tachycardic at 145 bpm, hypotensive with a BP of 90/50 mmHg. Physical exam revealed generalized abdominal tenderness, with guarding and rebound tenderness. Blood tests were ordered in accordance to the local guidelines. The results were significant for leukocytosis (18,500 white blood cells per cubic centimeter) and anemia (hemoglobin 8.6 mg/dL). His plain erect chest X-ray
showed free air in the peritoneum. As part of his acute management, the patient was kept nil by mouth with septic management pathway activated with IV fluid resuscitation and IV broad-spectrum antibiotics. The pneumoperitoneum in the erect plain CXR and his clinical picture, led to preliminary diagnosis of generalized peritonitis, indicating an emergency laparotomy. The ileum was perforated with associated purulent fluid in the peritoneal cavity. Affected part of the ileum was surgically resected and anastomosis was performed. This was followed by the abdominal cavity wash out with 10 L of sterile and warm saline. No further exploration was done at this stage, as the source of infection was identified to be the perforation and there was limited access to peritoneal cavity and exploration opportunity, due to the significant adhesions caused by previous surgeries. Additionally, surgical time had to be minimised due to patient's clinical instability.

The surgery was performed by a consultant general surgeon, with 5 years of experience and substantial expertise in colorectal cases, with the aid of two surgical residents. After the surgical procedure, the patient was transferred to the intensive care unit and had close observation and monitoring. Unfortunately, the patient remained hemodynamically unstable. While in the ICU and intubated, he received broad antibiotics (Imipenem, Tazocin, Vancomycin), antifungal medication (Caspofungin), continuous norepinephrine infusion, as well as fentanyl for pain and sedation, along with continuous maintenance IV fluid therapy. He had a cardiac arrest 36 h later, and was resuscitated successful with CPR and more intensive fluid resuscitation. Following this event, the patient turned the corner and showed clinical improvements in terms of vitals and urine output; given his trajectory of recovery he was extubated on day 5 post-op.

On day 7, the patient's recovery took an unexpected turn, with him deteriorating clinically and becoming febrile, once again. A triple (IV, oral, enema) contrasted abdominal CT was organized, to identify the source of fever. The CT scan (seen in Fig. 1) was remarkable for a large collection in the spleen containing gas and oral contrast. These radiologic finding was consistent with a colo-splenic fistula. In the theater, a large communicating orifice was found between the splenic flexure of the colon and spleen, shown in Fig. 2, with indications for splenectomy and left hemicolectomy. Pathology and histology report of the specimen revealed moderately differentiated adenocarcinoma in splenic flexure of the colon, with extensive necrosis with foci of viable adenocarcinoma in the spleen. The patient had an uneventful recovery after operation, with return of intestinal peristalsis, increase in appetite, and resolved septic status. The patient was discharged 40 days post-op. He was followed up regularly at one-month, three-month, and six-month by the surgical team as an outpatient, with patient remaining well and returning to his pre-morbid status. At his annual visit, his surveillance colonoscopy showed presence of ulcerated mass in the right colon, with the biopsy confirming adenocarcinoma requiring total colectomy and end ileostomy insertion. The patient was managed through a multidisciplinary team involving oncologist, radiotherapist, surgeon and his local family doctor as well as a counsellor.

3. Discussion

Internal fistulisation of the colon to other hallow viscera, such as the bladder, vagina, or small bowel is a relatively common and well documented in the literature, however colosplenic fistulas are extremely rare with approximately less than 20 reported cases worldwide [1,2]. The first case was reported by Naschitz et al. who documented colosplenic fistula formation in a patient with colonic lymphoma [2]. Similar to the more common fistulas, colosplenic fistulas may arise from inflammatory and neoplastic changes. These changes often associate with diverticulitis, Crohn's disease, pancreatitis, locally advanced colorectal cancer, lymphoma, melanoma, and as a complication of non-operative management of splenic trauma. In most of these cases, development of colosplenic fistulas associate with severe and poorly controlled Crohn’s disease [2-3,5-12].

The management of colosplenic fistulas are not well established in the setting of colorectal cancer. In most reported cases, the patients were managed surgically with splenectomy and left hemicolectomy. Review of the literature revealed only two documented cases with nonsurgical management: one with colosplenic fistulation on a background of pancreatitis and splenic hematoma, and the other case had a colosplenic fistula due to splenic infarct as a complication of polycythemia vera, fortunately both patients had favorable outcomes [9,11]. Here we need to emphasize that in these two cases, the primary pathology originated in the spleen subsequently causing erosion into the colon, unlike other cases where the initial pathology was in the colon. Based on the surgical management guidelines of Crohn's disease in the presence of sepsis, the patient should be treated with appropriate resuscitation and broad-spectrum antibiotics and abscess drainage, with persistent sepsis warranting resection of involved segment [13].

4. Conclusion

Colosplenic fistula is an exceedingly rare condition, often associated with malignancy or chronic inflammatory process in the gastrointestinal tract. As with any chronic inflammatory process, early detection of the disease process is necessary to prevent such complications and initiate early management. Diagnosis may be difficult due to the non-specific nature of the symptoms, yet early suspicion and considering CT scan are helpful in establishing an early diagnosis. Once diagnosed, the preferred approach to management consists of resuscitation, broad-spectrum antibiotics, and en-bloc resection of the affected portions of the colon and the spleen. This case highlights the importance of early initiation of genetic testing in young patients with early presentation of rectal adenocarcinoma, and serious consideration of a total colo-

![Fig. 1. Transverse cuts of abdominopelvic CT scan revealed a large collection in the spleen containing gas and oral contrast due to colo-splenic fistulisation.](image-url)
proctectomy, to prevent the iatrogenic burden of multiple invasive intervention on the patients and financial burden on the healthcare system. Furthermore, in young patients with colon cancer, colectomy may be advisable to prevent further malignancy development, however this is not the current recommended guidelines for rectal cancer. This would require further comparative observational research to provide evidence on the prognosis and outcome of patients with rectal cancer whom receive colectomy and those who receive alternative management options.

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Ethical approval

An emergent treatment done for a rare case of colosplenic fistula and after the successful treatment we reported the patient condition and the management, it was a retrospective study (case report) and there is no need for ethical approval, so we don’t have any ethical approval.

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.

CRediT authorship contribution statement

MA, surgeon, First Author, study conceptualization.
NM, surgeon, corresponding author, study conceptualization, edit of manuscript.
ZK, background research, literature review and writing up the case report.
ZA, Medical student, doing background research, edit of manuscript.

Declaration of competing interest

All authors decline any financial and personal relationships with other people or organisations that could inappropriately influence (bias) their work.

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Research registration

This case report is not a ‘First in Man’ report and don’t need registration to the registry website.

Guarantor

Narjes Mohammadzadeh.

References

[1] J.B. Goldberg, R.A. Moses, S.D. Holubar, Colosplenic fistula: a highly unusual colonic fistula, J. Gastrointest. Surg. 16 (12) (2012) 2338–2340, https://doi.org/10.1007/s11605-012-2033-0.
[2] J.E. Naschitz, D. Yeshurun, I.L. Horovitz, A. Dahaan, N.B. Lazaro, Y.E. Bos, Spontaneous colosplenic fistula complicating immunoblastic lymphoma, Dis. Colon Rectum 29 (8) (1986) 521–523, https://doi.org/10.1007/bf02262610.
[3] E. Pappalardo, A. Ricci, X. Dray, P. Marteau, P. Valleur, Splenic abscess secondary to a colosplenic fistula in Crohn’s disease, Acta Chir. Belg. 107 (3) (2007) 323–324, https://doi.org/10.1080/00015458.2007.11680066.
[4] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, A. Thoma, et al., The SCARE 2020 guidelines: updating consensus surgical consensus surgical case report (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
[5] L.R. Diana, F.L. George, Colosplenic fistula and splenic abscess complicating Crohn’s colitis, J. Clin. Gastroenterol. 21 (1) (1995) 74–75. https://pubmed.ncbi.nlm.nih.gov/7560839/.
[6] A. Gervaise, Roman C. De Saint, P. Sockeel, M. Lapierre, H. Darbois, et al., Splenic abscess secondary to a colosplenic fistula as the presenting manifestation of colon cancer, J. Radiol. 91 (12 Pt 1) (2010) 1259, https://doi.org/10.1016/s0221-0363(10)70184-0.
[7] A.A. Al-Zahir, A.W. Meshikhes, Colonic lymphoma presenting acutely with perforated colo-splenic fistula, Int. J. Surg. Case Rep. 3 (8) (2012) 368–371, https://doi.org/10.1016/j.ijscr.2012.08.013.
[8] M.S. Karpel Jr., D.G. Hicks, M.H. Torosian, Colon invasion by primary splenic lymphoma: a case report and review of the literature, Surgery 111 (2) (1992) 224. https://pubmed.ncbi.nlm.nih.gov/1736393/.
[9] P.J. Goldsmith, T.C. Demos, E.R. Guynor, Colosplenic fistula in a patient treated with Interleukin-2 for malignant melanoma, J. Comput. Assist. Tomogr. 21 (4) (1997) 674–676, https://doi.org/10.1097/00004728-199707000-00003.

Fig. 2. (2A) View of colo-splenic Fistula during Surgery in situ. (2B) Opened fistula is shown on the medial side of spleen (white Arrows).
[11] J.R. Means, E.R. Villella, K.R. Stahlfeld, Splenocolic fistula in a patient with polycythemia vera, Am. J. Surg. 185 (2) (2003) 173–174, https://doi.org/10.1016/S0002-9610(02)01210-2.

[12] S. McCrystal, M. Hatzifotis, Splenocolonic fistula following non-operative management of splenic rupture, Trauma 15 (1) (2013) 86–90, https://doi.org/10.1177/2F1460408612458737.

[13] S. Strong, S.R. Steele, M. Boutrous, L. Bordineau, J. Chun, D.B. Stewart, et al., Clinical practice guideline for the surgical management of Crohn’s disease, Dis. Colon Rectum 58 (11) (2015) 1021–1036. https://pubmed.ncbi.nlm.nih.gov/26445174/.