



Colosplenic fistula diagnosis and management: a case series and review of literature

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Background: A colosplenic fistula (CsF) is an extremely rare complication. Its diagnosis and management remain poorly understood, owing to its infrequent incidence. Our objective was to systematically review the etiology, clinical features, diagnosis, management, and prognosis to help clinicians gain a better understanding of this unusual complication and provide aid if it is to be encountered.

Methods: A systematic review of studies reporting CsF diagnosis in Ovid MEDLINE, Ovid EMBASE, Scopus, Web of Science, and Wiley Cochrane Library from 1946 to June 2022. Additionally, a retrospective review of four cases at our institution were included. Cases were evaluated for patient characteristics (age, sex, and comorbidities), CsF characteristics including causes, symptoms at presentation, diagnosis approach, management approach, pathology findings, intraoperative complications, postoperative complications, 30-day mortality, and prognosis were collected.

Results: Thirty patients with CsFs were analyzed, including four cases at our institution and 26 single-case reports. Most of the patients were male (70%), with a median age of 56 years. The most common etiologies were colonic lymphoma (30%) and colorectal carcinoma (17%). Computed tomography (CT) was commonly used for diagnosis (90%). Approximately 87% of patients underwent a surgical intervention, most commonly segmental resection (81%) of the affected colon and splenectomy (77%). Nineteen patients were initially managed surgically, and 12 patients were initially managed nonoperatively. However, 11 of the nonoperative patients ultimately required surgery due to unresolved symptoms. The rate of postoperative complications was (17%). Symptoms resolved with surgical intervention in 25 (83%) patients. Only one patient (3%) had had postoperative mortality.

Conclusions: Our review of 30 cases worldwide is the largest in literature. CsFs are predominantly complications of neoplastic processes. CsF may be successfully and safely treated with splenectomy and resection of the affected colon, with a low rate of postoperative complications.

Keywords: colosplenic fistula, splenocolic fistula, splenocolonic fistula, surgery, systematic review

Introduction

A colosplenic fistula (CsF) is an abnormal tract formed between the colon and the spleen. This extremely rare complication was first described in the 1980s^[1] but remains relatively poorly understood due to its infrequent occurrence. It has been previously observed in

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HIGHLIGHTS

- The largest review of rare colosplenic fistula (CsF) cases, including 30 patients internationally.
- Summary of CsF causes, clinical features, diagnosis, and management - Surgical intervention was the most successful treatment for symptom resolution.
- Colonoscopy caused no complications in the evaluation of CsF.

patients with neoplastic diseases, infections, inflammatory processes, trauma, congenital disorders, and even as a side effect of immunotherapy^[2–26]. However, its etiology remains difficult to understand, as all literature consists of single-patient case reports. Additionally, with only a limited number of CsF cases reported worldwide^[27], the diagnosis and management are vague. Hence, we performed a systematic review of patients with CsFs to summarize the etiologies, clinical features, diagnosis, management, and prognosis to help clinicians gain a better understanding of this unusual complication and provide aid if it were to be encountered.

Material and methods

A predesigned protocol was developed for this study. This study was reported according to the Preferred Reporting Items for Systematic

Reviews and Meta-Analysis (PRISMA, Supplemental Digital Content 1, <http://links.lww.com/JS9/B806>)^[28] and quality assessment was performed according to AMSTAR^[29] (Supplemental Digital Content 2, <http://links.lww.com/JS9/B807>) (Fig. 1). This review was registered in PROSPERO.

Eligibility criteria

For the review, we included studies reporting patients diagnosed with CsF, regardless of etiology or management.

Data sources and searches

We applied a search strategy developed in collaboration with an experienced librarian to find potentially eligible studies in Ovid MEDLINE, Ovid EMBASE, Scopus, Web of Science, and Wiley Cochrane Library from each database’s inception until

June 2022. A review of the literature was performed using the search terms ‘colosplenic fistula’, ‘splenocolic fistula’, and ‘splenocolonic fistula’. No language restrictions were imposed. Conference abstracts were included, and literature reviews were excluded. The reference lists of selected studies were searched to identify additional publications (Supplementary Search Strategy, Supplemental Digital Content 3, <http://links.lww.com/JS9/B808>).

The Cleveland Clinic Foundation – IRB 08-670 was used to conduct a retrospective review of the available CsF cases at our institution. Potential patients were identified from the electronic medical records using natural language processing and searching the operative reports of all surgeries related to using the search terms ‘colosplenic fistula’, ‘splenocolic fistula’, and ‘splenocolonic fistula’.

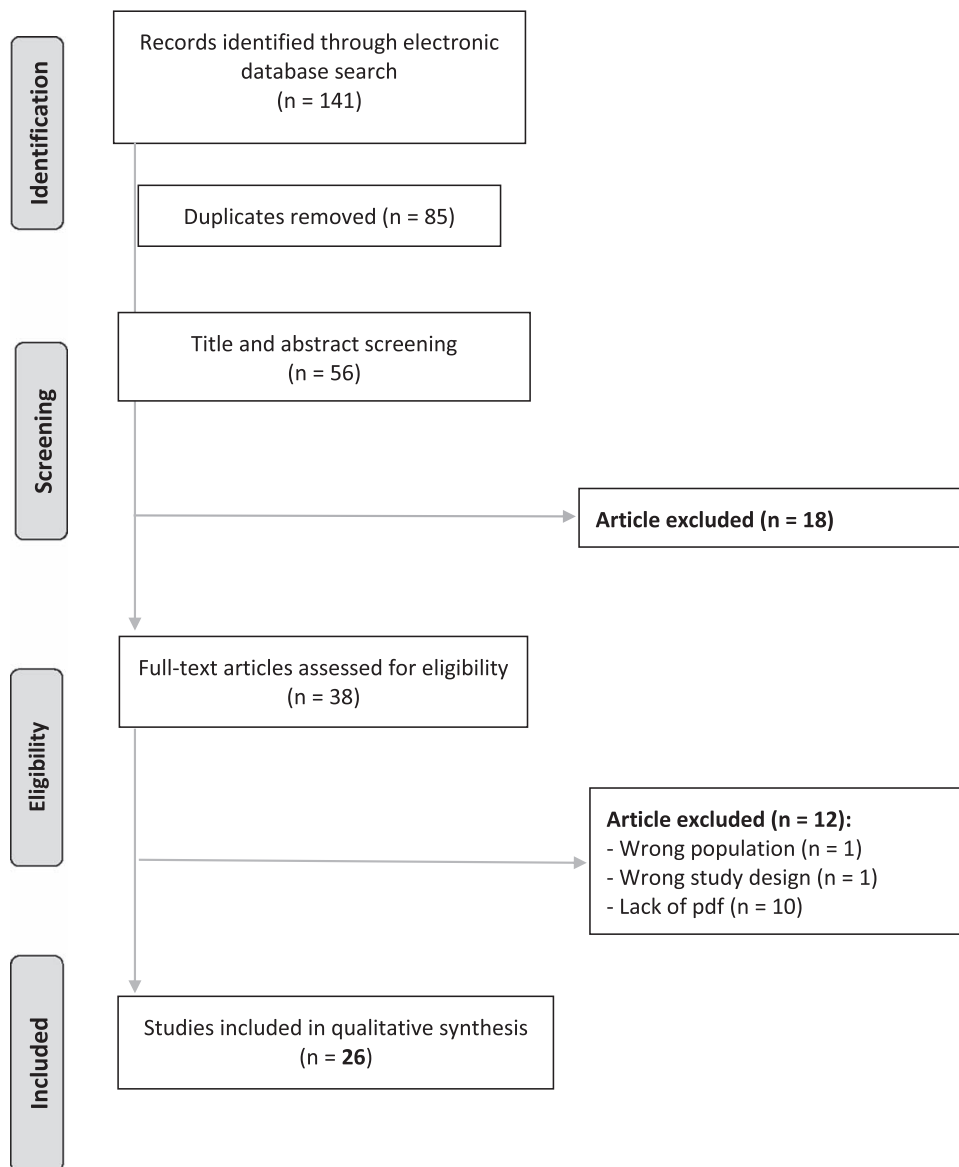


Figure 1. Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) flow diagram of studies selection process. An evidence-based set of items used to screen article titles, abstracts and full-texts to assess for eligibility in our scoping review.

Study selection

The search strategy results were uploaded to an online software program (Covidence Systematic Review Software; Veritas Health Innovation)^[30]. Next, the abstracts and full texts were screened. The reviewers (R.S. and F.A.C.) worked independently and in duplicate. Before initiating the study selection process, a pilot study with five articles was conducted to clarify the eligibility criteria. For abstract screening, articles included by at least one reviewer were considered for a full-text review. In the full-text review phase, only articles included by both reviewers were deemed eligible for this systematic review and in the case of disagreements, these were resolved by consensus with a third reviewer (E.P.L.). Full-text review agreement was substantial (Cohen's kappa = 0.7). Articles written in languages other than English or Spanish were translated using Google Translate (GT).

Data collection

Data from the included studies were extracted independently and in duplicate in a standardized manner by two reviewers (R.S. and F.A.C.). A third review checked all the included studies for accuracy (E.P.L.). The following information was extracted: general characteristics (first author's last name, publication date, and country), patient characteristics (age, sex, and comorbidities), CsF data collected included cause, symptoms at presentation, diagnosis approach, management approach, pathology findings, intraoperative complications, postoperative complications (Clavien–Dindo and 30-day mortality), and prognosis.

Methodological quality in individual studies

We used a tool proposed by Murad *et al.*^[31] to evaluate the methodological quality of the case reports. This tool is composed of four domains assessed by eight questions: (1) selection, (2) ascertainment, (3) causality, and (4) reporting. Case reports were assessed by two reviewers (E.P.L. and O.H.D.), with disagreements resolved by consensus with a third reviewer (T.U.). When only two or fewer questions were not satisfied, the risk of bias was considered low. Studies with three unsatisfied questions were assessed as unclear risk. Case reports with four or more questions not satisfied were considered to have a high risk of bias (Supplementary Table A.2, Supplemental Digital Content 4, <http://links.lww.com/JS9/B809>).

Statistical analysis

A narrative synthesis of the included studies was provided. We used descriptive statistics (e.g. percentages) to report our findings and summarized them in figures and tables.

Open data

To support reproducible research, we made all our files freely available on an online platform. Documents used in the screening phases (references screened in abstract screening and full-text screening) and extra information (tables and figures) are available at <https://figshare.com/s/4130858d368e899525ec>.

Results

The search strategy retrieved 141 references, of which 26 studies were included^[1–3,5–26] (Fig. 1). All the studies were case reports.

The exclusion criteria are summarized in Figure 1. The studies originated predominantly from the United States (43%) (Supplementary Table A.1, Supplemental Digital Content 5, <http://links.lww.com/JS9/B810>), and the overall risk of bias was judged to be unclear in eight studies and high in 18 studies. The latter was primarily attributable to a lack of sufficient detail, which would have allowed other investigators to replicate the research (Supplementary Table A.2, Supplemental Digital Content 4, <http://links.lww.com/JS9/B809>). Four patients were identified at our institution and were included in our analysis (Table 1).

We identified 30 patients with CsF. There were 21 (70%) males with a median age of 56 years (range, 2–84 years). Two pediatric cases were included: a 2-year-old female^[10], and a 16-year-old male^[9]. Twenty (67%) patients had comorbidities. Many patients presented with abdominal pain (76.7%), fever (50%), and lower gastrointestinal bleeding (27%). Other presenting symptoms included nausea/vomiting (23%), anemia (20%), and hemodynamic instability (13%). The most common causes of CsF were colon lymphoma (30%), colorectal adenocarcinoma (17%)^[2,3,5–10,14,23–25], Crohn's disease^[1,6–8] (13%), and infection (10%). The most common diagnostic modalities used to assess CsF was a computed tomography (CT) scan (90%), in which enteral contrast could be seen entering the spleen in seven cases. Colonoscopy was performed in seven (20%) cases without complications. CXR were completed in seven cases (20%) with low diagnostic utility. In four cases, a fistulogram was performed through the percutaneous drain showing a CsF. Initial management included surgery (63%), medical therapy without procedures or surgery^[1,8,11,13] (20%), and percutaneous drain^[5,15] (17%). One patient was transitioned to palliative care without any medical or surgical intervention. Of the patients initially managed nonoperatively, three of the six (50%) of the patient initially managed with medical therapy required surgical intervention to resolve symptoms. Symptoms resolved with surgery included fever (100%), lower gastrointestinal bleeding (100%), sepsis (100%), and abdominal pain (92%). Additionally, four of the five (80%) cases initially managed with percutaneous drain required surgical intervention due to unresolved septicemia (50%), increasing abdominal pain (50%), and moderate uncontrolled enteric output from drain (25%). Two of these four (50%) patients initially managed by percutaneous drain did not have worsening symptoms, but underwent definitive surgical management after a drain contrast-injection study showed a fistulous connection to the bowel. A total of 26 patients (87%) underwent surgical treatment which most commonly included a segmental colon resection in 21 patients (81%), and splenectomy in 20 patients (77%) (Table 2).

There were two cases that reported intraoperative complications, both from our case series. Complications included coagulopathy and hypothermia requiring urgent packing, open abdomen, and urgent transfer to the ICU. This was further complicated by missed gastrostomy that required reoperation with primary repair. The other intraoperative complication, a rectal wall injury, resulted during the colostomy reversal and was repaired primarily without further problems. There were five^[17,20,22] (17%) postoperative complications, which consisted of respiratory insufficiency (Clavien–Dindo I) (13%) and reoperation for missed gastrostomy (Clavien–Dindo IV) (3%). On follow-up, three (10%) of the 30 patients had a 30-day mortality. One patient developed multiple intra-abdominal abscesses

Table 1**Summary of colosplenic fistula cases at our institution.**

Age/sex	Comorbidities	Cause	Presentation	Diagnosis approach	Pathology	Management approach	Complications	Follow-up
Neoplasia								
55F	AIDS, Non-Hodgkin Lymphoma,	Non-Hodgkin Lymphoma	Nausea, fever, weight loss, fatigue, abdominal pain, lower extremity edema	CT – splenic abscess, colosplenic fistula Gastrograffin enema: fistula EGD: Normal Colonoscopy: normal	Diffuse Large B-cell lymphoma involving spleen and colonic wall	1' Laparotomy, segmental colectomy, end-colostomy, splenectomy, antibiotics 2' takedown of colostomy and primary anastomosis	Post-op: Respiratory insufficiency Intra-op: None	Mortality: No
77M	No	Non-Hodgkin's lymphoma	Nausea, vomiting, LUQ abdominal pain, fever	CT – splenic abscess, colosplenic fistula CXR: Left pleural effusion	Diffuse Large B-cell lymphoma	1' Laparotomy, splenic flexure colon resection, end-colostomy, splenectomy, antibiotics	Post-op: None Intra-op: None	Mortality: Yes, palliative care
69M	Arrhythmia, hypertension	Non-Hodgkin's lymphoma	Fever, abdominal pain, weight loss	CT – splenic abscess CXR: left pleural effusion	Diffuse Large B-cell lymphoma	1' Laparotomy, splenic flexure resection, splenectomy, antibiotics, open abdomen, abdominal packing 2' abdominal closure, transverse colostomy 3' abdominal re-exploration, gastrostomy repair 4' colostomy reversal	Post-op: Septic shock, respiratory insufficiency, return to OR for missed gastrostomy, pancreatic leak Intra-op: 1' coagulopathy, hypothermia, open abdomen with packing. Missed gastrostomy	Mortality: No
Miscellaneous								
76M	Non-Hodgkin's lymphoma, Stroke, CAD, infective endocarditis	Unclear	Bowel obstruction	CT – splenic abscess, colonic fistula	No Lymphoma	1' Laparotomy, segmental colon resection, end-colostomy, splenectomy, antibiotics	None	Mortality: No

AIDS, acquired immunodeficiency syndrome; CT, computed tomography; CXR, chest X-Ray; EGD, esophagogastroduodenoscopy; F, female; LUQ, left upper quadrant; M, male.

Table 2
Summary of colosplenic fistula review.

Variable/Group	Patients (n = 30)
Sex	
Male	21 (70.0%)
Age in years, median (range)	56 (2–84)
Comorbidities	20 (66.7%)
Presenting clinical features	
Abdominal pain	23 (76.7%)
Fever	15 (50.0%)
Leukocytosis	11 (36.7%)
LGIB	8 (26.7%)
Nausea/Vomiting	7 (23.3%)
Anemia	6 (20.0%)
Elevated CRP	6 (20.0%)
Hemodynamic instability/Shock	4 (13.3%)
Mechanism of fistula	
Lymphoma	9 (30.0%)
Colorectal adenocarcinoma	5 (16.7%)
Crohn's disease	4 (13.3%)
Infectious	3 (10.0%)
Trauma	1 (3.3%)
Pancreatic adenocarcinoma	1 (3.3%)
Other/Unknown	8 (26.7%)
Diagnostic modality performed	
CT Scan	27 (90.0%)
Enteral contrast in spleen on CT	7 (23.3%)
Colonoscopy	7 (23.3%)
CXR	7 (23.3%)
Contrast enema	5 (16.7%)
Abdominal US	4 (13.3%)
Fistulogram	4 (13.3%)
EGD	4 (13.3%)
Abdominal X-Ray	3 (10.0%)
Initial management	
Surgery	19 (63.3%)
Medical (No procedures or surgery)	6 (20.0%)
Percutaneous drain	5 (16.7%)
No Intervention/Palliative	1 (3.3%)
Transition to surgical management	(n = 11)
From initial medical group to surgery	3 of 6 (50.0%)
From percutaneous drain group to surgery	4 of 5 (80.0%)
Surgical procedures included	(26 patients, 86.7%)
Segmental colon resection	21 of 26 (80.8%)
Splenectomy	20 of 26 (76.9%)
Complications	
Clavien–Dindo I	4 (13.3%)
Clavien–Dindo IV	1 (3.3%)
Intraoperative complications	2 of 26 (7.6%)
Follow-up	
Mortality	3 (10.0%)
Related to surgery	1 (3.3%)
Unrelated to surgery (Progression of disease) with transition to palliative care	2 (6.7%)

CRP, C-reactive protein; CT, computed tomography; CXR, chest X-Ray; EGD, esophagogastroduodenoscopy; LGIB, Lower gastrointestinal bleed; US, ultrasound.

leading to septic shock, multiorgan failure, and died shortly following a laparotomy, segmental colon resection, and inferior pole splenectomy^[22]. One patient case at our institution successfully underwent laparotomy with splenic flexure colon resection, end colostomy, and splenectomy to treat the CsF but succumbed to progression of his primary malignancy. The other mortality underwent no intervention, was transitioned to comfort care, and

her death was due to the progression of the primary malignancy^[20]. (Table 1 and 2, and Supplementary Table A.1, Supplemental Digital Content 5, <http://links.lww.com/JS9/B810>).

Discussion

In this review, we summarize the etiologies, clinical features, diagnosis, management, and prognosis of 30 patients reported worldwide. We found that most CsFs were formed as a complication of neoplastic processes and Crohn's disease. In many cases, diagnosis is achieved using a CT scan with enteral and/or intravenous contrast. Most CsFs are treated with laparotomy, splenectomy, and resection of the involved colon. Finally, the rates of postoperative complications and mortality associated with CsF were (17 and 10%, respectively). Our pooled analysis of the available colosplenic cases may aid in the diagnosis and management of this rare entity.

Clinical features

Most patients with CsF were middle-aged males. The presenting symptoms vary depending on the underlying etiology. Nevertheless, several similar clinical features were observed in this small patient pool. About three-quarters of the patients reported pain or discomfort in the left upper quadrant of the abdomen, which may suggest location of pathology. However, a palpable left flank mass was rarely present and seems to not be an indicator of CsF. Notably, approximately a quarter of the cases reviewed reported LGIB, which is thought to be from the direct evacuation of the splenic pulp into the colon via the fistula^[10,13,26]. Anemia was also repeatedly seen, which could be due LGIB or chronic disease from an underlying malignancy. Another common feature was fever, leukocytosis, and elevated CRP to suggest an inflammatory process or infection. Unfortunately, CsF can present with hemodynamic instability and septic shock requiring urgent intervention.

Etiology and pathology findings

CsFs were first described in a 1985 case report as a rare complication of Crohn's disease^[1]. Subsequent case reports have identified CsFs in other contexts such as colonic lymphoma and colorectal cancer. Our review of available cases identifies colonic lymphoma as one of the most common causes of CsF. Similarly, in our institution, three of the four CsF cases were also diagnosed with colonic lymphoma. Interestingly, the gastrointestinal (GI) tract is the most common extranodal site of lymphoma, although the colon is rarely the site of involvement^[32–34]. Although less than 3% of colonic lymphomas perforate^[35], our analysis suggest colonic lymphoma perforations at the splenic flexure may lead to the formation CsF. Naschitz *et al.*^[25] hypothesized that colonic lymphomas perforate and form fistulas due to the massive tumorous involvement of tissues, extensive coagulative necrosis, and absence of desmoplastic reaction. This hypothesis is supported by several cases in our review, which found histological evidence of colonic lymphoma cells along the malignant fistulous tract and inside the spleen. Evidence from our review suggests that in the rare case of left colonic lymphoma, clinicians should be wary of CsFs as a complication.

CsFs have also been repeatedly observed as a complication of colon cancer of the splenic flexure. The cases reviewed in this

study reported fistula formation mostly because of malignancy-driven perforation at the splenic flexure, rather than from direct invasion into the spleen. Colon cancer perforation is either due to tumor necrosis at the cancer site, or to distended colon blowout from a distal malignant obstruction^[36]. Gervaise *et al.*^[22] postulated that after perforation of the colon at the splenic flexure, the local inflammatory process and adhesion formation trigger the development of a fistulous tract with the spleen. Although splenic flexure tumors only account for 3–5% of all colon cancer cases^[37,38], CsFs may be a complication of tumors at this location.

Crohn's is another cause of CsF. Crohn's disease causes chronic transmural inflammation throughout the gastrointestinal tract. Up to 50% of patients with Crohn's disease develop fistulas when this inflammation causes a transmural ulcer that penetrates from the affected bowel to adjacent structures^[39]. The majority of intra-abdominal fistulae are entero-enteric^[40]. However, our review revealed that patients with Crohn's disease can form a fistula from their diseased colon to the adjacent spleen. Many of these cases have reported deep ulcerations in the colon adjacent to the fistula, supporting Crohn's colitis as the etiology^[7,8].

Several cases of CsF have unique etiologies but a similar proposed mechanism of fistula formation. Two case reports suggested that a massive spleen can cause pressure necrosis in the adjacent colon and result in a fistula^[4,13]. Others have reported infectious causes of fistula development. For example, drainage from splenic abscess^[4,5] or echinococcal hydatid cyst^[26]. Similarly, our institution reported a case of endocarditis resulting in a splenic abscess and ultimately a CsF.

Diagnostic modalities

Several imaging modalities and endoscopic tools have been used to evaluate and diagnose CsF. In our review, chest radiography and abdominal plain radiography rarely offered diagnostic value. Fistulograms were effective under special circumstances in which the spleen was intubated with a percutaneous drain. In such cases, the contrast injected through the tube was shown to flow into the colon. Contrast enemas were also successful in showing contrast flow through the fistula and did not require percutaneous access. Enemas can be used as a confirmatory adjunct after noncontrast CT or splenic ultrasound raised suspicion of the fistula. Abdominal or splenic ultrasound (US) is helpful in identifying an enlarged spleen, splenic lesions, or air in the spleen. However, they have limited utility for the diagnosis of CsF. CT imaging either effectively diagnosed CsF or was highly suggestive based on the identification of a splenic abscess. CT with intravenous contrast helped identify splenic lesions and colonic thickening, whereas enteral contrast was useful in identifying the fistula tract as several cases showed enteral contrast entering the spleen. MRI was not performed in any case. A systematic review by Panes *et al.*^[41] suggested that CT, US, and MRI have high accuracy (sensitivity and specificity > 80%) in identifying intra-abdominal fistulas, but the selection between MRI and US is dependent on local expertise and availability. Colonoscopy was helpful in identifying the underlying etiology, but seldom identified a CsF. Snell^[2] suggested that colonoscopy had the potential to cause splenic rupture via insufflation through a CsF. However, several cases in this review performed colonoscopies without incident and therefore appears to be safe in the setting of CsF^[11–13,21,25,26].

Management

As most patients presented with fever and leukocytosis, they were empirically treated with intravenous (IV) fluid and intravenous broad-spectrum antibiotics. Depending on the clinical severity of the presenting symptoms or etiology, patients were either managed nonoperatively or with upfront surgery. Nonoperative approaches consist of addressing the splenic collection/abscess, which most typically involves placing an image-guided percutaneous drain into the spleen. However, this approach remained inadequate in addressing the fistula, as only one of the five cases did not require surgical treatment for resolution of symptoms. Reasons for failure included continued fever, worsening abdominal pain, continued septicemia, and uncontrolled feculent drainage from the drain. Therefore, percutaneous drains can help with diagnosis and temporizing measures, but have lower curative potential. Similarly, medical therapy without invasive procedure or surgery was helpful in stabilizing patients, but half eventually required surgical intervention.

Surgical management of CsFs usually includes open splenectomy and segmental resection of the involved colon. Undergoing both in conjunction seems to adequately address the fistula from both ends of the tract. One case attempted just an inferior resection of the spleen at the site of the fistula, but this patient died from complications of the surgery. Although only one case of this partial splenectomy was found, it appears that a full splenectomy is safer and effective in treatment of CsF. Additionally, due to the high incidence of neoplasia and Crohn's as cause of CsF, it is recommended that a segmental resection be performed. Although, if a neoplastic process is present, the extent of colon and spleen resection should be evaluated and aligned with patient's wishes and oncological team. For example, there was a case in which the etiology of CsF was advanced and surgical intervention would cause more harm than benefit. The patient was transitioned to comfort care and died shortly after from their malignancy^[20].

Prognosis

Overall, patients presenting with symptoms resolved after intervention to address CsF. Relatively few complications were reported in the nonoperative group and in those that received surgical intervention. Almost all postoperative recoveries were uneventful and showed resolution of CsF symptoms on follow-up. As expected, resolution of CsF occurred with surgical resection of the affected organs. Of the 30 cases reviewed, we identified two patients with intraoperative complications and five patients with postoperative complications. Four of these cases had relatively insignificant Clavien–Dindo I complications, including respiratory insufficiency and atrial fibrillation. However, one patient at our institution had Clavien–Dindo IV postoperative complications that required reoperation and intensive care management for missed gastrostomy. In the three mortalities, two of the cases had advanced underlying malignancy with progression prompting palliative care. Only one case of mortality could be possible related to the surgical treatment. This was a surgery in a patient with splenic flexure colon cancer complicated by perforation, CsF, and splenic abscess^[22]. In this case, there was an attempt at partial splenectomy of the inferior portion of the spleen.

Strengths and limitations

The main limitation of our review was the paucity of data regarding CsFs, which may reflect the true rarity of this complication or simply the lack of publication on the topic. In addition, some studies lacked granular data on the outcomes of interest. We were unable to statistically compare or analyze the outcomes. Finally, for the abovementioned reasons, we remain limited in our ability to draw strong recommendations, as we could only generate an overview of what is happening with patients with CsF. Therefore, this piece can serve as an educational tool.

Despite these limitations, the strengths of this study were notable. To our knowledge, this is the first global perspective systematic review describing the spectrum related to the care of patients with CsFs. Second, our inclusion of conference abstracts and the lack of language restrictions enabled us to include a satisfactory number of patients. Lastly, we followed a systematic methodology by applying prespecified and detailed data tabulation and extraction and standardized evaluation of evidence quality and publication bias. All steps were rigorously performed by multiple researchers.

Conclusions

In this study, we report the largest available review of CsFs. From the 30 identified cases worldwide, we concluded that CsFs were mainly formed as a complication of neoplastic processes or Crohn's disease. CT imaging is the most common modality used to identify the fistulous connection between the colon and spleen. Finally, we found that a surgical approach, including resection of the involved colon and splenectomy, had a low rate of post-operative complications and successful resolution of symptoms.

Ethical approval

Ethical approval for this study was provided by the Institutional Review Board of Cleveland Clinic Foundation, Cleveland, Ohio in the USA. IRB 08-670.

Consent

Written informed consent was obtained from the patient for publication and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

E.P.L., O.H.D., and S.D.H.: conceptualization; E.P.L., S.D.H., C.P., O.H.D., F.A.C., R.S., A.S., and T.U.: data curation; E.P.L., S.D.H., C.P., O.H.D., T.U., F.A.C., R.S., A.S., T.L.H., and S.R.S.: formal analysis; E.P.L., S.D.H., C.P., O.H.D., T.U., F.A.C., and R.S.: investigation; E.P.L., O.H.D., and S.D.H.: methodology; E.P.L., O.H.D., and S.D.H.: project administration; O.H.D., E.P.L., S.D.H., T.H., and S.R.S.: visualization; E.P.L., O.H.D., C.P., and T.U.: writing original draft; E.P.L., S.D.H., C.P., O.H.D.,

A.S., S.R.S., T.H., T.U.: writing – review and editing. All authors have read and approved the final manuscript.

Conflicts of interest disclosure

No conflicting interest to report. SDH: consultant fees, Shionogi, Takeda, Guidepoint. EPL: none OHD: none CP: none RS: none TU: None AS: none FAC: none TLH: none SRS: none.

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Guarantor

Stefan D. Holubar.

Data availability statement

To support reproducible research, we have made all our files freely available in an online platform. Documents used in the screening phases (references screened in abstract screening and full-text screening) and extra information (tables, figures) are available in this site: <https://figshare.com/s/4130858d368e899525ec>.

Provenance and peer review

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Disclaimers

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Assistance with the study

Not applicable.

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