

Spontaneous Bilateral Lumbar Region Colocutaneous Fistulae. A Case Report

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Summary

Bilateral spontaneous lumbar colocutaneous fistulae are rare. They are associated with high costs of care, morbidity, and mortality. We present the case of a 50-year-old male patient who presented with a 10-year history of two low-output spontaneous colocutaneous fistulae from both the left and right lower lumbar regions with no identifiable risk factors. He was managed operatively with segmental left colon resection and a temporary loop ileostomy with uneventful recovery after reversal of the ileostomy 2 months later. Whether the etiology of the fistulae is clinically apparent or not, the management should follow commonly known principles for enterocutaneous fistula care. This report highlights a case of bilateral lumbar region spontaneous

colocutaneous fistulae of unknown cause that was managed operatively with satisfactory short-term outcomes.

Keywords: Spontaneous, Colocutaneous, Fistula, Kenya, Case report

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Introduction

Entero-cutaneous fistulae (ECFs) generally result from iatrogenic causes, malignancy, inflammatory bowel disease, trauma, or rarely occur spontaneously. Some can also result from complicated hernias (1, 2). Most, however, result after surgery due to the breakdown of anastomoses or inadvertent bowel injuries (3). Fistulae are associated with high morbidity and mortality. They equally attract high healthcare costs, pose a huge burden to patients psychologically, and offer a challenging problem to surgeons in terms of care and operative repair (4, 5).

Spontaneous enterocutaneous fistulae account for approximately 10–25% of ECFs (6). Common risk factors include colonic diverticulitis, malignancies, radiation, inflammatory conditions (e.g., inflammatory

bowel disease), infections, and malignancies. Other causes include mesenteric ischemia (3). However, the majority are caused by diverticular disease and malignancy (7). They can present as a wound discharging enteric contents, with pain and tenderness, distension, or complications that might include sepsis, malnutrition, and electrolyte imbalances (8). While a proportion of spontaneous fistulae will close with nonoperative management alone, surgery is a key component in their treatment. The choice between surgery and nonsurgical management largely depends on patient factors, pathology, and the specific course of the disease (5, 8, 9).

Spontaneous colocutaneous fistulae are a rare sub entity (3, 7), and bilateral ones are rarer. There are few reports

in the literature from Africa on their incidence and management. In this report, we present a rare and unusual case of spontaneous bilateral lumbar region colocutaneous fistulae from Kenya, which we opine to be the first case to be reported from Africa.

Case presentation

Presentation

A 50-year-old male patient from Nakuru, Kenya, presented with a 10-year history of feculent drainage from two wounds on the lumbar regions, bilaterally. They had started as skin indurations with localized pain and itchiness, first occurring on the left followed by the right, approximately 3 months apart. They soon developed into abscesses that ruptured spontaneously, resulting in fistulae that he managed with local dressings only. The daily drainage from the wound was less than 20 cc as reported and was mainly composed of formed semi-solid stool. The right wound had ceased drainage a month prior to presentation. Interestingly, he had at no point sought care during the 10 years. He gave no specific reasons.

He did not have any identifiable cause, suggestive evidence or risk factors for immunosuppression and did not have any prior surgeries. He also did not have a history of preceding trauma or prior exposure to radiation and lacked symptoms of, or previous diagnosis or treatment for tuberculosis, inflammatory bowel disease, or cancer. He had no history of melena stools, hematochezia, or constipation. He had no history of alcohol intake or smoking. During the 10 years, he had not used any prescription medications aimed at treating the wounds or the drainage.

He was not wasted and had a body mass index of 26 kg/m². He was afebrile and did not have conjunctival pallor and neither did he have pedal edema. On examination of the abdomen, he had a 1-cm wound on the right lumbar region, 8 cm superior to the iliac crest on the midaxillary line, (Figure 1) and a 2-cm wound on the left lumbar region 6 cm superior to the iliac crest around the midaxillary line with soiling of the surrounding skin (Figure 2). The fistulae had surrounding thick tethered scar tissue and hyperpigmentation of the skin. He lacked prior surgical

scars or organomegaly. There were no anorectal lesions noted on digital rectal examination. The rest of his systemic examination was normal.



Figure 1. Fistula on right lumbar region with surrounding thick tethered scar tissue and hyperpigmentation of the skin.



Figure 2. Fistula on left lumbar region with surrounding thick tethered scar tissue and hyperpigmentation and soiling of nearby skin.

His pre-operative complete blood count parameters, electrolytes, and liver function tests were within normal, as shown in Table 1. A contrast computed tomography (CT) scan of the abdomen revealed two colocutaneous fistulae from both the right and left colon with localized

inflammation around the left fistula tract. (Figure 3) abscess or distal obstruction. There was no radiological evidence of intra-abdominal

Table 1. The peri-operative laboratory parameters

PARAMETER	VALUE (UNITS)		NORMAL RANGES
	PRE-OPERATIVELY	POST-OPERATIVELY	
White cell count	6.5 (*10 ⁹ /L)	4.0 (*10 ⁹ /L)	4–11
Granulocytes	66.1(%)	66.0 (%)	40–70
Lymphocytes	8.7 (%)	9.0 (%)	20–40
Hemoglobin	12 (g/dL)	11(g/dL)	12–18
Hematocrit	37.3 (%)	36 (%)	36.1–50.3
Platelets	417 (*10 ⁹ /L)	390 (*10 ⁹ /L)	150–400
MCV	69.3 (fl)	69.3 (fl)	80–97
MCH	29.4 (pg)	28.2 (pg)	26.7–33.7
MCHC	32.8 (g/dL)	30 (g/dL)	32.7–25.5
Albumin	43 (g/L)	40 (g/L)	40–50
Creatinine	0.9 (mg/dL)	1.01 (mg/dL)	0.7–1.35
Urea	7.1 (mmol/L)	8 (mmol/L)	1.73–8.3
Potassium	4.1 (mEq/L)	3.6 (mEq/L)	3.6–5.5
Sodium	136 (mEq/L)	130 (mEq/L)	135–150
Chloride	111 (mEq/L)	112 (mEq/L)	97–108
Fasting blood sugar	4.7 (mmol/L)	–	3.9–5.6
Random blood sugar	5.4 (mmol/L)	6.0 (mmol/L)	4.6–7.8
HIV status	Negative		

Management

After careful optimization and a multidisciplinary review involving nutritionists, radiologists, physicians, a wound care team, a psychologist, and the surgical team, he was scheduled for operative management. An enema was given on the night before the day of surgery and the last liquid meal given 3 hours before the scheduled operation time. One gram of ceftriaxone was administered preoperatively, one hour before the incision was made, according to our institutional protocol. Under general anesthesia via a midline abdominal entry, we found a large fistulous tract from the distal descending colon to the skin with a large intervening cavity containing devitalized tissues. On the right side, after medial reflection of the cecum and

ascending colon, fibrotic adhesions of the cecum and anterior abdominal wall were found with no obvious fistula- signs of a spontaneously healed fistula. Nothing further was done here, but we fashioned a diverting loop ileostomy 3 ft proximal to the ileocecal junction.

A segmental left colon resection was performed with primary end-to-end anastomosis. The cavity was debrided both transabdominally and from the external fistula. The right-sided sinuses were taken down, debrided off the devitalized tissues, and left open.

We started enteral feeds with normal diet on day 2 after surgery. He received IV Ceftriaxone 1 g twice daily and IV metronidazole 500 mg thrice daily for 4 days post-operatively. He was also put on IM morphine and IM Ketorolac for analgesia. The recovery was uneventful,

and he was discharged on the 6th post-operative day. His midline incision healed well with the debrided fistulae undergoing serial wound care.

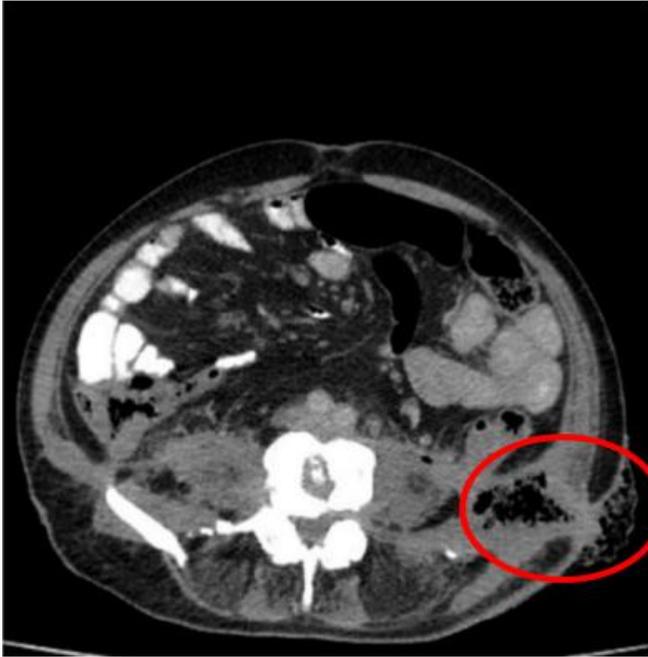


Figure 3. Fistulous tract from left colon with localized surrounding inflammatory changes (marked with a circle). The right side shows no obvious fistulous.

The ileostomy was taken down at 8 weeks. He developed superficial surgical site infection at the ileostomy site on the 2nd week post the take-down. Thorough local debridement resolved the infection with subsequent delayed primary closure.

Histology of resected segment was negative for malignancy. It revealed a fistulous tract with mucosal ulceration, active chronic non-granulomatous inflammation, and stromal hemorrhage (*Report included as Supplementary file*).

He has since fully recovered and has resumed his activities of daily living. He was scheduled for a colonoscopy within the following year, in accordance with the national screening guidelines for colorectal cancer.

Discussion

The most common causes of spontaneous colocutaneous fistulae include colonic diverticula, malignancy,

radiation, inflammatory bowel disease, specifically Crohn's disease, and infections like tuberculosis and amoebiasis (10). Immunosuppressed patients are also at a particularly higher risk (11). The patient we report was neither immunosuppressed nor did he have any identifiable risk factor for spontaneous fistulae.

Spontaneous colocutaneous fistulae are very rare globally (12, 13). Nagaraja and colleagues opine that this could be due to the lack of population-based studies (13). In fact, there are just a few reported cases of spontaneous colocutaneous fistulae from Africa. This is despite the high prevalence of tuberculosis in this region and the rising incidence of colorectal tumors (14). In Africa, unlike in high-income regions, spontaneous colocutaneous fistulae caused by diverticular disease is expected to be minimal as the incidence of diverticular disease in this population is low (15). Most reported cases of spontaneous colocutaneous fistulae are unilateral and single, with no cases of bilateral or multiple spontaneous fistulae (1, 2, 7).

The management of colocutaneous fistulae is hinged on eradication of sepsis, nutritional optimization, including fluid and electrolyte repletion and delineation of the fistula anatomy (4, 5, 7). This is best done in the setting of a multidisciplinary team (16). Specifically, control of sepsis involves resuscitation, antibiotic therapy according to local protocols, drainage of intra-abdominal collections, and wound care (17). Wound care should be focused on containing the effluent, improving comfort and skin protection. Enteral feeding is preferred where possible. However, the route and quantity of feeds should be goal-oriented and individualized (17, 18). In addition to fluid repletion, reducing the fistula output is a key strategy to reduce loss of fluids and electrolytes. This can be achieved by reducing or restricting intake of hypo-osmolar fluids, use of antisecretory and antimotility agents, and consideration of bowel rest and parenteral nutrition (19). Subsequently, the pathology underlying the fistula is defined. This is followed by proposal of a procedure to surgically treat the fistula if conservative management is deemed unsuccessful or inapplicable (9).

A fistulogram, a barium enema, and a CT scan can be utilized to study the fistula anatomy, that is, to detect the

site of the fistula and delineate the fistula tract. CT scan can, however, provide additional details on the associated pathology and is usually the initial modality in practice. We therefore favored a CT scan as opposed to a fistulogram in the work-up of our patient (3, 9). Other modalities include magnetic resonance imaging (MRI), MR fistulography, and endoscopy. Indeed, a colonoscopy can help identify an occasional malignancy and contribute in the assessment for distal obstruction (12). Our patient did not have a colonoscopy during the peri-operative period and histology from fistula site was negative for malignancy- this informed the decision to book him for a colonoscopy in the following year after surgery.

Colocutaneous fistulae are classified as Sitzes-Serra type III enterocutaneous fistulas (large bowel fistula without a large abdominal wall defect) and can be managed conservatively in most cases (7). Haack et al. note that the rate of spontaneous closure of spontaneous fistulae ranges from 5% to 20% and is influenced by the underlying pathology and the ability to correct it (5). Importantly, infection and chronic inflammation can lead to protracted fistula closure (4). Despite our patient having a low-output colocutaneous fistula, his fistulae took a protracted time to heal. We did not consider other conservative strategies, such as the use of fibrin sealants, endoscopic clips, or fistula plugs, largely due to the unavailability of the devices. Although the fistula on the right eventually healed spontaneously, the active inflammation on the left, as revealed by the pathology results, may have impeded healing and led to the chronicity. In such instances, resection of the diseased bowel segment at the fistula site is usually recommended to avoid further complications (7). This should be accompanied by anastomosis of the healthy bowel ends (5). Simple fistulectomy with primary closure of the bowel wall defect is associated with high recurrences (20). Since the right colon fistula had spontaneously healed and was not active, we fashioned a diverting ileostomy to rest the proximal right colon, protect the anastomosis on the left, and avoid a second anastomosis. Operative management aims at avoiding creation of new enterotomies and re-establishing bowel continuity (18). For example, Singh et al. successfully managed a patient

with spontaneous colocutaneous fistula at the cecal area following tuberculosis with right hemicolectomy and ileo-transverse anastomosis, along anti-tuberculous drugs (20). Although no guidelines exist on the best timing for surgical management, when contemplated, it should be delayed for at least 12 weeks (9, 21). The healing rate after operative management approaches 90% (22). However, the surgeon should realize that the outcomes can also be disappointing (21).

Limitations

Our main limitation was a short follow-up period to the writing of this report. However, we enrolled the patient on a prolonged follow-up owing to his yet unidentified probable risk factor(s). In addition, performing a pre-operative colonoscopy and a stool analysis would have perhaps added vital information in the management of our patient.

Conclusion

Bilateral lumbar region spontaneous colocutaneous fistulae are rare. A fistulogram, CT scan of the abdomen and/or pelvis, and colonoscopy should be considered in the work-up of patients with such fistulae. This report highlights a case of bilateral lumbar region spontaneous colocutaneous fistulae without clinically identifiable etiology that was managed operatively with regard to the widely known principles for ECF care with satisfactory short-term outcomes.

Ethical consideration

Our patient gave a written informed consent for publication of this report.

Author contributions

All authors contributed equally in the conceptualization and writing of the first draft to reviewing and editing the original draft.

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