Original Article

Enterovesical Fistulae: Case Series Report from a Single Institution

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Key Words

Colovesical fistula; Colorectal cancer; Diverticulitis; Colectomy **Purpose.** Enterovesical fistula is an uncommon disease that reduces the quality of life. A fistula can be chronic and induces severe sepsis which results in mortality. The etiology of fistula can be a complicated inflammation or malignancy. The medical condition of the patient is always complex. We would like to review cases of colovesical fistula to improve the treatment and care.

Methods. Patients who been discharged from the colorectal surgery department of Taipei Veteran General Hospital from 2004 to 2018 were reviewed. 46 patients with colovesical fistula were confirmed after the medical chart review. We reviewed their demographics, notes, image reports, and laboratory data.

Results. The median age was 68 years. The most common etiology was diverticulitis (37.0%). Malignancy had 34.8% and radiation had 19.6%. CT scan established 25 colovesical fistula diagnosis (54.3%). All cases who received open and laparoscopic one-stage surgery were cured without recurrence. Only one in eleven cases who had diversion alone end up closing the stoma successfully.

Conclusions. The colovesical fistula or rectovesical fistula usually happens in patients with advanced malignancy. One-stage surgery was optimal to cure fistulae. Two-stage surgery with a protective ostomy was considered for complicated cases. Fistula repair or collectomy should be done to treat colovesical fistula or rectovesical fistula.

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Enterovesical fistula (EVF) is an abnormal connection between the intestine and urinary bladder. It is a rare diagnosis in the surgical department, accounting for approximately one of every 3000 surgical admissions.¹ It occurs mostly in the sigmoid colon and bladder dome.² This study would like to focus on colonvesical fistula (CVF) and rectovesical fistula (RVF). The most common etiology of CVFs is complicated diverticulitis.^{3,4} Cancer and Crohn's disease are also common etiologies.^{2,5} Men are more predis-

posed to having CVFs than women, with a male-tofemale ratio of approximately 2-3:1.^{6,7} The symptoms of CVF can persist for many years, having a significant impact on the quality of life. It could also cause severe urosepsis, which could lead to multi-organ failure.^{2,6} The most common clinical presentations are pneumaturia, fecaluria, dysuria, and frequency.^{8,9} In this study, we present a case series of CVF and RVF patients received over a 15-year period in a single institution.

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Materials and Methods

The cases were enrolled retrospectively from the discharge database of the Division of Colon and Rectal Surgery, Taipei Veteran General Hospital, from January 2004 to December 2018. The keywords "fistula" and "bladder" (or "vesical") were used to search the discharge notes of all discharged patients. Those with anal fistula or vaginal fistula were excluded. In addition, patients were excluded if information about their chief complaint or diagnostic process was missing in the discharge note, progress note or admission note. In total, 79 patients were confirmed to have CVF or RVF according to their history, chief complaint, and imaging findings. Their admission notes, progress notes, discharge notes, operative notes, nursing records, and imaging studies were reviewed to extract information about their chief complaint, symptoms, diagnostic process, treatment, and outcome. The diagnoses were made according to diagnostic criteria for diverticulitis, cancer, or other causal histories, together with at least one clinical symptom of EVF. The image evaluation included computed tomography (CT), magnetic resonance imaging (MRI), colonoscopy, cystoscopy, and cystography. These studies confirmed the diagnosis in their final reports. Direct identification in the operative specimen was also one of the confirmation methods.

Thirty-three EVF patients were excluded from the study as follows: 23 patients who had fistula as a complication of a previous colectomy, 7 patients who could not undergo any definite treatment because they had a critical or terminal condition such as hollow organ perforation, tumor bleeding, cachexia or multiple metastases, 2 patients did not come back for their scheduled surgery after diagnosis, and one patient was instead confirmed to have a prostate fistula in the end. The 23 colectomy-related patients had limited choice of surgical treatments due to the difficulty of repeat colectomy. This group of patients had different surgical treatment concern from the others so they were excluded. Finally, 46 patients were enrolled in the current case series.

In this study, statistical analysis was done using the R and Excel software. Student's t-test and Chisquare test were used to analyze the quantitative and categorical data, respectively. A *p*-value less than 0.05 was considered to be statistically significant.

Results

From 2004 to 2018, a total of 46 patients (27 males, 19 females) were diagnosed with CVF or RVF, accounting for 2 in every 1000 admissions in the Division of Colon and Rectal Surgery. The median age of fistula diagnosis was 68 years with a range of 28-92 years. The median follow-up duration was 786 days (IQR, 325 to 1712).

The most common etiology was diverticulitis (37.0%), while malignancy accounted for 34.8% of the cases. Among the patients with a malignancy etiology, the most common malignancy was colorectal cancer (30.4%) while other cancers were prostate (2.2%) and bladder cancers (2.2%). In 9 other patients who also had a malignancy, the etiology of their fistulas was radiation-related (19.6%) and not due to tumor invasion. Other less common etiologies were genito-urinary iatrogenic perforation, trauma, and Crohn's disease (Table 1). The most common investigation for fistula identification was computed tomography (CT) (Table 2), which confirmed the diagnosis of 25 CVFs

Table 1. Etiology	of enterovesical	fistula in this series
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	Number	%
Diverticulitis	17	37.0
Malignancy	16	34.8
Sigmoid colon cancer	10	
Rectal cancer	3	
Cecal cancer	1	
Prostate cancer	1	
Bladder cancer	1	
Radiation	9	19.6
Cervical cancer	7	
Prostate cancer	1	
Rectal cancer	1	
Other	4	8.7
GU iatrogenic perforation	2	
Trauma	1	
Crohn's disease	1	

Data are number of patientsand the percentage

GU, genitourinary surgery

or RVFs (54.3%). The other imaging studies like colonoscopy, MRI, cystoscopy, and cystography were also useful tools for CVF and RVF identification (Table 2). Three patients had their fistulae identified during surgery (6.5%). Five patients had only a clinical diagnosis based on symptoms of fecaluria, pneumaturia or micturition from the anus (10.9%). Among the clinical symptoms, fecaluria was the most common complaints (45.7%) (Table 3). The median interval from symptoms to diagnosis was 1 month (IQR, 0 to 4).

Three of the patients had rectal cancer and they underwent a two-stage surgery, involving low anterior resection with protective colostomy, followed by reversal of the colostomy. One of them had colostomy closure without recurrence. Another 56-year-old man had a rectovesical fistula due to rectal cancer recurrence, presenting with fecaluria after colostomy closure and was treated with a colostomy and an ileal conduit. In the last case, colostomy closure was not achieved due to uncured anastomosis leakage.

In ten patients, EVF was due to sigmoid colon cancer. Among them, one underwent laparoscopic anterior resection, five underwent open anterior resection, and two underwent a two-stage operation including anterior resection and colostomy. There was no recurrence in these eight patients. One 78-year-old woman had sigmoid colon cancer with liver metastasis. She first underwent colostomy and was discharged uneventfully. However, she died due to unknown reasons one month after the diagnosis outside the hospital. Another 80-year-old man with liver metastasis first underwent a colostomy but developed persistent urosepsis. He underwent another surgery with anterior resection and transureteroureterostomy; however,

Table 2. Initial diagnostic method for enterovesical fistulas

	Number	%
СТ	25	54.3
Colonoscopy	4	8.7
MRI	4	8.7
Cystoscopy	2	4.3
Cystography	3	6.5
Operation finding	3	6.5
Not identified	5	10.9

Data are number of patients and the percentage.

the sepsis still persisted after the operation and the patient died.

One patient had prostate cancer patient and underwent an abdominoperineal resection, while another had cecal cancer and successfully underwent an anterior resection with right hemicolectomy. An 80-yearold man with bladder cancer due to bladder cancer recurrence in his neobladder with liver metastasis underwent a colostomy. However, he died due to persistent urosepsis after the colostomy procedure.

Seventeen patients had diverticulitis of which, three cases underwent laparoscopic anterior resection without postoperative complications and seven cases had open anterior resection. One of the seven cases was an 82-year-old man who developed jejunum entero-cutaneous fistula after the surgery. He died due to leakage after the fistula repair. Six of the seventeen cases underwent a two-stage operation involving anterior resection and colostomy. Four cases had successful colostomy closure, while one case did not have colostomy closure due to rectal stricture. Another 74year-old woman with end-stage renal disease had fistula recurrence and developed septic arthritis. She underwent another surgery involving hip resection, sigmoidectomy, and fistula repair. However, she died due to urosepsis. A three-stage operation was performed in one patient who first required treatment for liver abscess before the colectomy. This case had no recurrence after colostomy closure.

Nine patients had CVF or RVF after radiotherapy without cancer recurrence. A two-stage operation was successfully performed in these two cases. The other seven cases had a permanent ostomy without repair.

Table 3. Patients' cli	nical symptoms
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	Number	%
Fecaluria	21	45.7
UTI	15	32.6
Abdominal pain	11	23.9
Pneumaturia	8	17.4
Dysuria	6	13.0
Urine from anus	5	10.9
Diarrhea	5	10.9
Hematuria	1	2.2

Data are number of patients and the percentage.

UTI, urinary tract infection.

Two of the seven cases still had urinary tract infection recurrence. One 92-year-old man died due to postoperative aspiration pneumonia.

A Crohn's disease patient successfully underwent a one-stage procedure involving ileocecal resection and sigmoidectomy. A trauma patient underwent rectovesical fistula repair and colostomy, but still had urine passage per anus. After the procedure was repeated, there was no more recurrence and the colostomy closed in the end. The iatrogenic fistulas were caused by transurethral resection of the prostate and open lithotripsy. One of them underwent sigmoidectomy and the other underwent a colostomy, although urine passage per anus still persisted. The colostomy was closed successfully after transanal repair.

The main treatment goal was to relieve the symptoms while the final aim was to treat the symptoms without an ostomy. The patients were assigned into two groups: those with and without an ostomy at their last follow-up. A total of 31 cases had no ostomy at their last follow-up (Table 4). The two groups were comparable with regards to median age (64 vs. 80 yr.; p = 0.0127), etiology, and operation method. More patients with diverticulitis were cured of the fistula (48.4 vs. 13.3%, p = 0.0474). Also, more radiation cases could not achieve ostomy closure (6.5 vs. 46.7% patients, p = 0.0047). Every patient who underwent open or laparoscopic one-stage surgery could be cured without an ostomy. Furthermore, those who had only ostomy creation, but no colectomy or fistula repair were less likely to be cured (1 vs. 10 patients, p <0.0001).

EVF could be a cause of death in some serious cases. Younger patients were more likely to survive for more than one year after a diagnosis of CVF or RVF (64.5 vs. 80 yr., p = 0.0144) (Table 5). Only stage IV patients survived for less than a year after fistula diagnosis in this study. Most patients received ostomy only in the poor survival group (17.5 vs. 66.7%, p = 0.0340).

Discussion

EVF can be a complication of diverticulitis and

colorectal cancer.² Diverticulosis is a common disease in Taiwan and colorectal cancer is the most common cancer in Taiwan.¹⁰ The symptoms of EVF like pneumaturia, lower urinary tract symptoms, and fecaluria can have a great impact on a patient's quality of life. Even though a colorectal surgeons may not be the first physician to make the diagnosis, we plays an important role in the management of the disease.

Diverticulitis and colorectal cancer accounted for most of the CVF and RVF etiology. G. Garcea et al. showed that CVF etiologies were diverticular disease (72.2%), colonic carcinoma (15.3%), and Crohn's disease (9.7%).⁶ Our series reported more colonic carcinoma and less Crohn's disease. Taiwan has a high colorectal cancer incidence; the age-standardized incidence rate in 2015 was 45.3 per 100,000 population.^{11,12} Taipei Veteran General Hospital is a referral medical

Table 4. Comparing the patients with and without ostomy at the last follow up

last tollow up			
	w/o	w/	
	ostomy at	ostomy at	
	last f/u	last f/u	p
	(n = 31),	(n = 15),	
	n (%)	n (%)	
Sex			
Man	20 (64.5)	7 (46.7)	0.4047
Age	64 (52.5-74)	80 (68-84)	0.0127
DM	6 (19.4)	3 (20.0)	1.0000
Etiology			0.0032
Diverticulitis	15 (48.4)	2 (13.3)	0.0474
Malignancy	10 (32.3)	6 (40.0)	0.852
Stage II CRC	6	0	0.1737
Stage III CRC	2	2	0.8271
Stage IV CRC	2	4	0.1495
Radiation	2 (6.5)	7 (46.7)	0.0047
Other	4 (12.9)	0	0.3693
Operation			0.0003
Open one-stage surgery	15	0	0.0032
Lapa. one-stage surgery	4	0	0.3693
Open Two-stage surgery	9	4	1.0000
Diversion alone	1	10	< 0.0001
Three-stage surgery	1	0	1.0000
Repaired + colostomy	1	0	1.0000
APR	0	1	0.7076

Data are number of patients with percentages in parentheses or median with a range in parentheses.

DM, diabetes mellitus; CRC, colorectal cancer; APR,

abdominal perineal resection; lapa., laparoscopic.

	Survive more than one yr. $(n = 40)$, n (%)	Survive less than one yr. $(n = 6)$, n (%)	р
Sex			
Man	23 (57.5)	4 (66.7)	1.0000
Age	64.5 (54.5-76.3)	80 (78.5-81.5)	0.0144
DM	7 (17.5)	2 (33.3)	0.7189
Etiology			0.0774
Diverticulitis	15 (37.5)	2 (33.3)	1.0000
Malignancy	13 (32.5)	3 (50.0)	0.7042
Stage II CRC	6	0	0.7133
Stage III CRC	4	0	0.9731
Stage IV CRC	3	3	0.0256
Radiation	8 (20.0)	1 (16.7)	1.0000
Other	4 (10.0)	0	0.9731
Operation			0.4552
Open one-stage surgery	14	1	0.6699
Lapa. one-stage surgery	4	0	0.9731
Open two-stage surgery	12	1	0.8491
Diversion alone	7	4	0.0340
Three-stage surgery	1	0	1.0000
Repaired + colostomy	1	0	1.0000
APR	1	0	1.0000

Table 5. Comparing the patients who survived and did not survive in one year

Data are number of patients with percentages in parentheses or median with a range in parentheses.

yr., year; DM, diabetes mellitus; CRC, colorectal cancer; APR, abdominal perineal resection.

center that receives the complicated colorectal cancer cases. Taiwan has far less Crohn's disease incidence than western countries; Crohn's disease incidence rate was 3.5 per 100,000 persons in 2013 according to a National health insurance research database-based study.^{13,14} This could explain the fact that this series only had one Crohn's disease patient.

CT is the gold standard imaging test for CVF and its diagnostic accuracy is approximately 90%-100%.^{15,16} This series did not repeat the same result because most of our CTs did not follow the instructions required for performing the scan before intravenous and after oral contrasts.¹⁵⁻¹⁷ Our CTs were usually done without oral contrast, and the intravenous contrast was usually administered before scanning. The findings on CT, which are suggestive of enterovesical fistulae include (1) air in the bladder (in the absence of previous lower urinary tract instrumentation), (2) oral contract medium in the bladder on nonintravenous contrast enhanced scans, (3) presence of colonic diverticula, and (4) bladder wall thickening adjacent to a loop of thickened intestine. Without oral contrast before scanning and intravenous contrast, we could not identify the fistula tract, which is a more direct evidence. In 8 patients (17.4%), surgery was done without any image identification. In this cases, the diagnosis of EVF is usually based on clinical symptoms like pneumaturia and micturition from the anus; therefore, the patients' history is also important. S.F. Najjar et al. suggested that the diagnostic verification of a CVF is necessary to identify the location of the fistula and guide the surgical plan.⁴ The author of this report had a different opinion; I believe that verification is not necessary when the etiology was already a surgical disease and the history and clinical symptoms were compatible with the diagnosis. However, this can only be proven by a trial.

This study excluded patients who underwent nonoperative treatment. These cases were excluded due to their foreseeable short life expectancy. They were in a terminal stage of malignancy or had severe sepsis without a chance of surviving from a general anesthesia operation. Few studies have showed the safety of non-operative management for selected patients.^{18,19} Those selected patients could not benefit from surgery. For patients who had multiple morbidities, unfit for general anesthesia, or foreseeable short life expectancy, conservative treatment may be an option. Conservative treatments includes NPO, urine diversion, and antibiotics. Most studies concluded on the necessity of surgery in all patients who are fit because of the high risk of septic complications and low self-healing rate.^{6,20,21}

Bertelson et al. reported that a one-stage procedure was effective for diverticular CVF in their 14 cases.²² Cochetti et al. also recommended treating CVF with a one-stage surgery if possible.²¹ The single-stage procedure is now the most common treatment of choice.^{6,21,23,24} They suggested that a multistage procedure should be performed only in patients with pelvic abscess formation, advanced malignancy or previous radiation therapy.²¹ Our series also revealed that radiation-related EVFs are more likely not to be healed. Seven of nine cases could not have their ostomy closed. Stage IV colorectal cancer demonstrated a poor prognosis; this is not only because of the malignancy but also because of persistent sepsis due to the unrepaired fistula. Levenback et al. preferred bowel resection with or without urinary conduit than isolation of the fistulized bowel loop and urinary conduit.²⁵ Diversion alone was also not recommended. In their series, one out of 3 were healed in the diversion group, 2 out of 4 in the isolation group, and 5 out of 6 in the resection group. Our series revealed the importance of fistula repair including colon resection. Diversion alone usually resulted in persistent sepsis and disease progression. One-stage surgery should be considered the first line treatment for the patients. In addition, laparoscopic surgery could be performed if the patient is eligible. Since younger and diverticulitis patients had better results, they may be good candidates for the one-stage surgery. On the other hand, a twostage surgery was also a proper option, especially for the elderly and those who received radiation. Diversion alone could be a cause of poor prognosis because the fistula was less likely to be cured.

The limitations of this study include the retrospective design and limited number of cases within the single institute. In addition, the treatment was based on the discretion of the attending physician rather than a consensus protocol.

Conclusions

CVF and RVF had favorable outcomes in young patients and in those having diverticulitis as etiology. Patients who underwent radiation therapy may need a permanent ostomy. Performing a one-stage surgery could cure CVF and RVF in selected patients. However, we should consider a two-stage surgery with protective ostomy if a repeat repair is inevitable. Finally, despite the limited experience, diversion alone might not be adequate for sepsis control and a definite surgical intervention should be timely considered to avoid persistent sepsis.

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<u>原 著</u>

腸膀胱廔管:單一機構病例系列報告

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目的 腸膀胱廔管是一個不常見的疾病但會嚴重影響生活品質, 屢管可以是慢性的也可以引發敗血症進而造成死亡, 屢管的病因可以是複雜的發炎或癌症, 我們將瀏覽這些大腸膀胱廔管的病例來精進廔管的治療。

方法 研究台北榮民總醫院大腸直腸外科 2004 到 2018 年出院的病人,有 46 個病人最後確診並收錄,我們瀏覽他們的基本資料、病例、影像報告、實驗室檢查。

結果 年齡的中位數是 68 歲,最常見的病因為憩室炎 37.0%,另外癌症 34.8%、放射治療 19.6%。有 54.3% 的病例可以由電腦斷層診斷出大腸直腸膀胱發炎。所有接受開腹與腹腔鏡一階段手術的病人都成功治療且沒有復發。11 個只單獨接受造口治療的病例,其中只有一位廔管癒合並成功關造口。

結論 大腸直腸膀胱廔管常發生在晚期惡性腫瘤。一階段手術是適當的治療,兩階段手術適合在較為複雜的病例上使用。大腸直腸膀胱廔管的理想治療應該要包含廔管修補或腸切除。

關鍵詞 大腸膀胱廔管、大腸直腸癌、憩室炎、大腸切除術。