Transanal endoscopic operation for rectocutaneous fistula after low anterior resection: a case report

Manuscript Number: INTSURG-D-20-00017R2

Full Title: Transanal endoscopic operation for rectocutaneous fistula after low anterior resection: a case report

Article Type: Case Report

Keywords: Anastomotic leakage; Enterocutaneous fistula; Low anterior resection; Transanal endoscopic operation; Rectocutaneous fistula; Case report

Corresponding Author: Ta-Wei Pu, MD
National Defense Medical Center
Taipei, TAIWAN

Corresponding Author Secondary Information:

Corresponding Author's Institution: National Defense Medical Center

Corresponding Author's Secondary Institution:

First Author: Yu-Jen Chen, MD

First Author Secondary Information:

Order of Authors:
Yu-Jen Chen, MD
Ta-Wei Pu, MD
Gang-Hua Lin, MD
Nung-Sheng Lin, MD
Jung-Cheng Kang, MD
Cheng-Wen Hsiao, MD
Chao-Yang Chen, MD
Je-Ming Hu, MD
Tzu-Chiao Lin, MD

Order of Authors Secondary Information:

Abstract:
Introduction: Enterocutaneous fistulas (ECFs) can be caused by abscess formation at the site of anastomotic leakage (AL) after surgery. Rectocutaneous fistula following low anterior resection (LAR) is rare, and medical management of ECFs is usually the initial treatment. We report a case of rectocutaneous fistula after laparoscopic LAR, which was successfully treated, for the first time, with a transanal endoscopic operation (TEO).

Case Presentation: A 58-year-old man presented with a history of hypertension, benign prostatic hyperplasia, peptic ulcer, and recent diagnosis of rectal cancer. The patient underwent laparoscopic LAR with coloanal anastomosis complicated with AL. He then underwent transanal repair of the anastomosis site and laparoscopy with ileostomy. Six months later, he complained of a painful mass lesion over the right buttock that relieved after passing purulent fluid and feces. Colonoscopy and imaging revealed a fistula for which he received antibiotics and wound incision and drainage. He also underwent TEO repair of the rectal fistula, recovered well, and was discharged from hospital. On follow-up 7 months later, there was no recurrence or sign of localized infection.

Conclusion: TEO repair may be an effective method for managing rectocutaneous fistula after LAR complicated with AL instead of a major operation.
Transanal endoscopic operation for rectocutaneous fistula after low anterior resection: a case report

Short title: Fistula treated by transanal endoscopic operation

Yu-Jen Chen, MD¹, Ta-Wei Pu, MD²,³,⁴,*, Gang-Hua Lin, MD¹, Nung-Sheng Lin, MD⁵, Jung-Cheng Kang, MD³, Cheng-Wen Hsiao, MD⁴, Chao-Yang Chen, MD⁴, Je-Ming Hu, MD⁴, Tzu-Chiao Lin, MD⁴

¹Department of Surgery, Tri-Service General Hospital, National Defense Medical Center, Taipei 114, Taiwan
²Department of Surgery, Division of Colon and Rectal Surgery, Songshan Branch, Tri-Service General Hospital, National Defense Medical Center, Taipei 105, Taiwan
³Department of Surgery, Division of Colon and Rectal Surgery, Taiwan Adventist Hospital, Taipei 105, Taiwan
⁴Department of Surgery, Division of Colon and Rectal Surgery, Tri-Service General Hospital, National Defense Medical Center, Taipei 114, Taiwan
⁵Department of Emergency Medicine, Armed Forces Taoyuan General Hospital, Taoyuan 325, Taiwan

Disclaimers: The authors declare that they have no conflicts of interest.

*Corresponding Author: Ta-Wei Pu, MD
Division of Colon and Rectal Surgery, Department of Surgery, Songshan Branch
Tri-Service General Hospital, National Defense Medical Center, Taipei 105, Taiwan
Tel: +886-2-27642151
Fax: +886-2-27642151
Email: tawei0131@gmail.com

**Sources of support:** None.

**Acknowledgments**
We thank Songshan Branch of Tri-Service General Hospital, National Defense Medical Center, Taipei, Taiwan.
Transanal endoscopic operation for rectocutaneous fistula after low anterior resection: a case report

ABSTRACT

Introduction: Enterocutaneous fistulas (ECFs) can be caused by abscess formation at the site of anastomotic leakage (AL) after surgery. Rectocutaneous fistula following low anterior resection (LAR) is rare, and medical management of ECFs is usually the initial treatment. We report a case of rectocutaneous fistula after laparoscopic LAR, which was successfully treated, for the first time, with a transanal endoscopic operation (TEO).

Case Presentation: A 58-year-old man presented with a history of hypertension, benign prostatic hyperplasia, peptic ulcer, and recent diagnosis of rectal cancer. The patient underwent laparoscopic LAR with coloanal anastomosis complicated with AL. He then underwent transanal repair of the anastomosis site and laparoscopy with ileostomy. Six months later, he complained of a painful mass lesion over the right buttock that relieved after passing purulent fluid and feces. Colonoscopy and imaging revealed a fistula for which he received antibiotics and wound incision and drainage. He also underwent TEO repair of the rectal fistula, recovered well, and was discharged from hospital. On follow-up 7 months later, there was no recurrence or sign of localized infection.

Conclusion: TEO repair may be an effective method for managing rectocutaneous fistula after LAR complicated with AL instead of a major operation.

Keywords: Anastomotic leakage; Enterocutaneous fistula; Low anterior resection; Transanal endoscopic operation; Rectocutaneous fistula
**INTRODUCTION**

Enterocutaneous fistulas (ECFs) are abnormal lesions caused by inflammatory bowel disease, surgery, abscesses, or iatrogenic causes such as abscess formation at the site of anastomotic leakage (AL). Non-operative closure rates for ECFs are lower than surgical closure rates. Transanal endoscopic operation (TEO) is a novel, minimally invasive surgical technique for treating rectal lesions. Although it is technically difficult to resect very large lesions, TEO can be performed in a limited space with parallel placement of the instruments. Here, we report the case of a 58-year-old man who presented with a painful mass lesion over the right buttock six months after having undergone laparoscopic low anterior resection (LAR) with coloanal anastomosis complicated with AL. We diagnosed a rectocutaneous fistula and treated him with a TEO.

**CASE PRESENTATION**

A 58-year-old Taiwanese man had a medical history of hypertension, benign prostatic hyperplasia, peptic ulcer, and recent diagnosis of rectal cancer. The patient underwent laparoscopic low anterior resection with coloanal anastomosis and the pathology report revealed adenocarcinoma of the rectum, moderately differentiated, invading to the pericolic fat, pT3N0M0, and stage IIA after surgery. Four days after the operation, the patient started complaining of abdominal pain. Diffuse abdominal tenderness was noted while the physical examination was performed. He then underwent transanal repair of the anastomosis site with V-lock and laparoscopy with ileostomy and lavage of the abdominal cavity and received broad-spectrum antibiotics for rectum anastomosis leakage.

Six months later, the patient complained of a painful mass lesion over the right buttock and experienced slight relief of the symptom after passing purulent fluid and feces. A low-grade fever of 37.5°C was noted while the physical examination revealed perineal tenderness.
Laboratory testing showed that the white blood cell count was $5.48 \times 10^3/\mu$L, with 85.5% neutrophil count and 8.3% lymphocyte count. The c-reactive protein level was 12.13 mg/dL. Colonoscopy (Figure 1) revealed a rectal fistula around the anastomosis site. An abdominal and pelvic CT scan (Figure 2A) revealed irregularly-shaped poorly-enhancing lesions in the right perineal region, suggestive of an infection, and a rectocutaneous fistula extending from the skin to the rectum (Figure 2B and 2C). The patient was treated with broad-spectrum antibiotics and received wound incision and drainage with a Penrose drain.

After six weeks, the patient underwent a TEO (Figure 3) for repair of the rectal fistula. A TEO surgery port (Transanal Endoscopic Operations, TEO, Karl Storz, Germany) was used on our patient. Surgery was performed with the patient placed in the Lloyd-Davies position under general anesthesia. After platform insertion, the pneumorectum pressure was set at 10–12 mmHg. A rectal fistula was noted (Figure 3A). Hook-type monopolar electrocautery was used for debridement and coagulation. The debridement was started on the necrotic tissue around the rectal fistula margins. The intraluminal pressure was reduced (7–8 mm Hg) to avoid increasing the tension during endoscopic suturing (Figure 3B). The full-thickness rectal fistula defect was closed with running sutures using the V-Loc wound closure device (Medtronic, Dublin, Ireland) (Figure 3C).

The patient recovered well and was discharged from the hospital after one week. After one month, revision of the ileostomy was performed smoothly and he recovered well and was discharged. Seven months after discharge, he had no signs of localized infection and no recurrence of the rectocutaneous fistula.

**Informed Consent**

Written informed consent was obtained from the patient for the publication of this report and the accompanying images.

**DISCUSSION**
ECFs are abnormal tracts that connect the alimentary system to the skin; the causes of ECFs include inflammatory bowel disease, surgery, abscesses, or iatrogenic causes.\textsuperscript{1} In our case, the reason for the rectocutaneous fistula formation might have been abscess formation at the site of the AL, which gradually caused weakening of the rectal wall and tissue necrosis.

Most cases of AL occur 3.5–8 days after performing LAR.\textsuperscript{4-6} A previous study showed that the incidence of colorectal AL increased in patients with tumors at the distal site (size larger than 3 cm) at an advanced stage or because of emergency surgery and metastatic disease.\textsuperscript{7} Intraoperative risk factors included blood loss/transfusion and duration of surgery of more than four hours.\textsuperscript{7} Another risk factor for fistula formation is the misusage of staplers.\textsuperscript{8} Currently, stapled anastomosis in colorectal surgery is becoming the “gold standard”.\textsuperscript{8} It improves the anastomotic technique and reduces bleeding; however, attention should be paid when firing the staple.\textsuperscript{8} In our case, there was no obvious risk factor for AL.

ECF treatment is focused on the control and treatment of sepsis, nutritional optimization, and anatomic mapping.\textsuperscript{1} The mortality and morbidity rates from ECFs have been reported to be between 6% and 33%.\textsuperscript{9} Medical management is usually the initial treatment. Nonoperative closure rates for ECF tracts are between 5% and 20%.\textsuperscript{2} Surgical closure rates for ECF tracts are around 75%–85%.\textsuperscript{2} Transanal endoscopic microsurgery (TEM), which was developed and defined by Prof. G. F. Buess, has generally been accepted for the treatment of early rectal cancers and benign rectal lesions but cannot be performed for colonoscopic excision.\textsuperscript{10-12} TEM involves a three-dimensional viewing system with a rectoscope and allows the creation of a pneumorectum, providing access to the entire rectum.\textsuperscript{13} However, because of its high cost, the learning curve, and the complexity of the equipment, TEM is not commonly used.\textsuperscript{14} In our case, exploratory laparotomy or laparoscopic management of rectocutaneous fistula could have been limited by intra-abdominal adhesions given the history of a previous operation. Hence, we opted to perform TEO on our patient. From the surgical viewpoint,
TEO appears to be as efficient as the classical TEM; TEO has shown to have a lower cost with the use of standard laparoscopic equipment and better ergonomics (owing to the camera).\(^{16}\)

In conclusion, the main advantage of TEO is that it enables surgeons to expose the lesion using endoscopy with a novel surgical technique. TEO also provides a better view than conventional transanal surgery to assess middle and upper rectal lesions. Finally, surgeons can accurately dissect or debride middle and upper rectal lesions. Thus, patients with fistula tracts that develop after LAR can avoid undergoing a major operation such as exploratory laparotomy or laparoscopic management of rectocutaneous fistula.
REFERENCES

1 Rahman FN, Stavas JM. Interventional radiologic management and treatment of enterocutaneous fistulae. J Vasc Interv Radiol. 2015 Jan;26(1):7-19; quiz 20. DOI: 10.1016/j.jvir.2014.09.009. Epub 2014 Nov 18.

2 Owen RM, Love TP, Perez SD, Srinivasan JK, Sharma J, Pollock JD, et al. Definitive surgical treatment of enterocutaneous fistula: outcomes of a 23-year experience. JAMA Surg. 2013 Feb;148(2):118-26. DOI: 10.1001/2013.jamasurg.153.

3 Han Y, He YG, Lin MB, Zhang HB, Lv KZ, Zhang YJ, et al. Treatment of preoperatively diagnosed colorectal adenomas by transanal endoscopic microsurgery: the experience in China. Asian J Endosc Surg. 2013 Aug;6(3):177-80. DOI: 10.1111/ases.12027. Epub 2013 Mar 20.

4 Floodeen H, Hallböök O, Rutegård J, Sjödahl R, Matthiessen P. Early and late symptomatic anastomotic leakage following low anterior resection of the rectum for cancer: are they different entities? Colorectal Dis. 2013 Mar;15(3):334-40. DOI: 10.1111/j.1463-1318.2012.03195.x.

5 Fouda E, El Nakeeb A, Magdy A, Hammad EA, Othman G, Farid M. Early detection of anastomotic leakage after elective low anterior resection. J Gastrointest Surg. 2011 Jan;15(1):137-44. DOI: 10.1007/s11605-010-1364-y. Epub 2010 Oct 27.

6 Maeda H, Okamoto K, Namikawa T, Akimori T, Kamioka N, Shiga M, et al. Rarity of late anastomotic leakage after low anterior resection of the rectum. Int J Colorectal Dis. 2015 Jun;30(6):831-4. DOI: 10.1007/s00384-015-2207-9. Epub 2015 Apr 11.

7 McDermott FD, Heeney A, Kelly ME, Steele RJ, Carlson GL, Winter DC. Systematic review of preoperative, intraoperative and postoperative risk factors for colorectal anastomotic leaks. Br J Surg. 2015 Apr;102(5):462-79. DOI: 10.1002/bjs.9697. Epub 2015 Feb 19.
8 Morarasu S, Frunza T, Rotundu A, Dimofte G, Lunca S. Fistulas after low anterior resection with TME. Jurnalul de Chirurgie. 2017 Jun;13(1):25-29. DOI: 10.7438/1584-9341-13-1-5

9 Williams LJ, Zolfaghari S, Boushey RP. Complications of enterocutaneous fistulas and their management. Clin Colon Rectal Surg. 2010 Sep;23(3):209-20. DOI: 10.1055/s-0030-1263062.

10 Casadesus D. Surgical resection of rectal adenoma: a rapid review. World J Gastroenterol. 2009 Aug 21;15(31):3851-4. DOI: 10.3748/wjg.15.3851.

11 Suppiah A, Maslekar S, Alabi A, Hartley JE, Monson JR. Transanal endoscopic microsurgery in early rectal cancer: time for a trial? Colorectal Dis. 2008 May;10(4):314-27; discussion 327-9. DOI: 10.1111/j.1463-1318.2007.01448.x. Epub 2008 Jan 10.

12 Bach SP, Hill J, Monson JR, Simson JN, Lane L, Merrie A, et al. Association of Coloproctology of Great Britain and Ireland Transanal Endoscopic Microsurgery (TEM) Collaboration. A predictive model for local recurrence after transanal endoscopic microsurgery for rectal cancer. Br J Surg. 2009 Mar;96(3):280-90. DOI: 10.1002/bjs.6456.

13 Bretagnol F, Merrie A, George B, Warren BF, Mortensen NJ. Local excision of rectal tumours by transanal endoscopic microsurgery. Br J Surg. 2007 May;94(5):627-33. DOI: 10.1002/bjs.5678.

14 Maslekar S, Pillinger SH, Sharma A, Taylor A, Monson JR. Cost analysis of transanal endoscopic microsurgery for rectal tumours. Colorectal Dis. 2007 Mar;9(3):229-34. DOI: 10.1111/j.1463-1318.2006.01132.x.

15 Serra-Aracil X, Mora-Lopez L, Alcantara-Moral M, Caro-Tarrago A, Navarro-Soto S. Transanal endoscopic microsurgery with 3-D (TEM) or high-definition 2-D transanal
endoscopic operation (TEO) for rectal tumors. A prospective, randomized clinical trial. Int J Colorectal Dis. 2014 May;29(5):605-10. DOI: 10.1007/s00384-014-1849-3. Epub 2014 Mar 28.

16 Nieuwenhuis DH, Draaisma WA, Verberne GH, van Overbeeke AJ, Consten EC. Transanal endoscopic operation for rectal lesions using two-dimensional visualization and standard endoscopic instruments: a prospective cohort study and comparison with the literature. Surg Endosc. 2009 Jan;23(1):80-6. DOI: 10.1007/s00464-008-9918-8. Epub 2008 Apr 29.
FIGURE LEGENDS

Figure 1. Colonoscopy: rectocutaneous fistula (white arrow) around the anastomosis site

Figure 2. Abdominal and pelvic computed tomography. A: irregularly-shaped poorly-enhancing lesions in the right perineal region, which might have been due to an infection (white arrow); B: a rectocutaneous fistula extending from the skin to the rectum, coronal view (white arrow); C: a rectocutaneous fistula extending from the skin to the rectum, axial view (white arrow).

Figure 3. Transanal endoscopic operation steps. A: The fistula (white arrow); B: Suture placement: the defect is closed using an endoscopic suture with a V-Loc device (white arrow); C: The fistula was closed by V-Loc sutures (white arrow).
