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# Entero-cuteneous fistula presenting as an insect bite: A case report Scripps Memorial Hospital La Jolla, Sharp Memorial Hospital San Diego, Des Moines College of Osteopathic Medicine

### INTRODUCTION

Enterocutaneous fistulas that develop between the colorectal anastomosis and the skin are usually a late manifestation of an anastomotic breakdown. This may be from an unrecognized leak, low grade ischemia at the anastomosis, or stricture. The prevalence of enterocutaneous fistulas in the general population is not known. There are very few reports in the literature of fistulas that include the gluteal region or lower limb.



### **OBJECTIVES**

- 1. To discuss the process by which an entero-cutaneous fistulas may form.
- 2. To present a case report of a woman with an enetrocutaneous fistula to the lower leg after TaTME
- 3. To discuss treatment and prevention options



Dordea et al. 2008

# **CASE REPORT**

A 61-year-old female presented to an outside emergency room complaining of a pain and swelling in her left popliteal fossa. She had a history of rectal cancer, status post transanal total mesorectal excision (TaTME) with neoadjuvant chemoradiation therapy and had been lost to follow-up for 1 year by her treating surgeon. At the time of her index operation, her lesion was noted at 4 cm from the anal verge and her post neoadjuvant staging was T0. Her pathology showed complete resolution of the cancer.

Upon new presentation, the patient complained of pain with ambulation and swelling over the past 3 months in her posterior thigh. She also complained of intermittent fevers and fatigue over the past 3 months. The patient thought she had been bitten by a spider and attributed her symptoms to an insect bite. She was diagnosed with an abscess, thought to represent infected papular urticaria from an insect bite. However, the abscess had ruptured and started draining what appeared to be stool combined with undigested food.



Physical examination of the involved thigh revealed a significantly larger posterior compartment than the contralateral leg, with thigh circumference being approximately 60 cm, whereas the uninvolved thigh circumference measured 40 cm. The posterior compartment of the thigh appeared erythematous and felt tense, with tenderness to deep palpation. Approximately 3 cm proximal to the popliteal fossa, a 1cm x 2cm ulcerated lesion was identified with surrounding erythema and induration, with both feculent and purulent drainage, as well as undigested food matter such as corn. The patient was unable to ambulate due to pain. No adenopathy or other lesions were identified. On digital rectal examination a severe anastomotic stricture was noted.

Laboratory studies revealed and elevated C-reactive protein (CRP) of 85 and erythrocyte sedimentation ratio (ESR) of 141, with a markedly elevated white blood cell (WBC) count of 32,000. The patient, however, was afebrile, normotensive, and had a normal resting heart rate.



Radiographs of the thigh were unremarkable. Computed tomography (CT) revealed: presacral fat infiltration and a 1.9 x 1.6 x 6.3 cm enhancing focus along the anterior aspect of the right sacrum and coccyx. There was a 3.6 cm rim-enhancing collection in the posterior left perirectal region, suspicious for an abscess. The lesion extended inferolaterally from this collection into the left gluteus maximus muscle. Gas and inflammatory changes in the distal left gluteus maximus muscle and in the posterior compartment of the left thigh, consistent with myositis and cellulitis were also described. There were multiple intramuscular abscesses within the distal biceps femoris measuring up to 2.2 cm as well.

Given her history of rectal cancer and radiation, her clinical picture created suspicion for a rectocutaneous fistula and infection of the posterior compartment of her thigh. As a result, the patient underwent colostomy with incision and debridement of the thigh to decompress the abscess and excise the fistula. She remained stable during her colorectal procedure, but upon decompression of the abscess, became suddenly hypotensive and tachycardic. The patient returned to the operating room for multiple debridement procedures before eventual delayed primary closure. Soft tissue biopsies were taken from the rectum and fistula, both negative for malignancy. Cultures taken from the fistula grew Enterococcus faecalis, Proteus mirabilis, and Candida albicans, treated with ampicillin-sulbactam with fluconazole for a course of 6 weeks.

The patient recovered well, but 4 months later re-fistulized, this time to her upper thigh, approximately 15 cm distal to her anus and 25 cm proximal to the original fistula.



Her drainage was limited to mucous isolated from the rectum. Due to concern for future infections, her nutritional status was optimized, and she ultimately underwent a robotic abdominoperineal resection with gluteal V-Y advancement flap and excision of the fistula. Biopsies again were negative for malignancy and cultures grew Proteus mirabilis, so she was treated with amoxicillin-clavulanate for 14 days, per infectious disease specialist recommendation. The patient recovered without incident or further fistula and at 2 years follow-up remained in good health.

Entero-cutaneous fistulas occur for numerous reasons. Most commonly from foreign body, history of radiation therapy, persistent infection (anastomotic leak), ischemia or inflammation, poor wound epithelization usually secondary to malnutrition, neoplasm, distal obstruction, and sepsis. The patient herein described had a previous history of rectal cancer, s/p TaTME (trans-anal total mesorectal excision) with prior neoadjuvant therapy. TaTME has been demonstrated to be safe and effective. Long term complications include conduit ischemia, anastomotic stricture, and fecal incontinence. However, long-term oncologic outcomes of TaTME have not been reported. This case study is interesting as the fistula developed likely due to conduit ischemia (low grade), the patient had some severe structuring at the anastomosis. Had this been an anastomotic leak, the patient would have presented with a posterior sacral abscess and would have been readily noticeable within the first few months of surgery. This patient also had the fistula track through the posterior thigh, rather than through the surrounding gluteus as is customary for fistulas associated with TaTME. This is remarkable that the patient did not show symptoms earlier or become septic.

Denost Q, Adam JP, Rullier A, Buscail E, Laurent C, Rullier E. Perineal transanal approach: a new standard for laparoscopic sphincter-saving resection in low rectal cancer, a randomized trial. Ann Surg. 2014;260(6):993-999. doi:10.1097/SLA.0000000000000766

Detering R, Roodbeen SX, van Oostendorp SE, et al. Three-Year Nationwide Experience with Transanal Total Mesorectal Excision for Rectal Cancer in the Netherlands: A Propensity Score-Matched Comparison with Conventional Laparoscopic Total Mesorectal Excision [published correction appears in J Am Coll Surg. 2020 Sep;231(3):411]. J Am Coll Surg. 2019;228(3):235-244.e1. doi:10.1016/j.jamcollsurg.2018.12.016

Dordea M, Venkatsubramaniam AK, Green SE, Varma JS. Delayed rectal anastomotic dehiscence presenting as a colocutaneous fistula in the popliteal fossa. Can J Surg. 2008;51(3):E65-E66

Drabble EH, Greatorex RA. Colocutaneous fistula between the sigmoid colon and popliteal fossa in diverticular disease. Br J Surg. 1994;81(11):1659. doi:10.1002/bjs.1800811133

Evaristo-Méndez G, Sánchez-Hernández AT, Melo-Velázquez A, Ventura-Sauceda FA, Sepúlveda-Castro RR. Fístula sigmoideo-glútea por diverticulitis: Reporte de una rara complicación [Sigmoido-buttock fistula by diverticulitis: report of a rare complication]. Cir Cir. 2013;81(2):158-162.

Garay Burdeos M, García-Botella M, Viciano Pascual V, et al. Postoperative radiotherapy-induced morbidity in rectal cancer. Rev Esp Enferm Dig. 2004;96(11):765-772. doi:10.4321/s1130-01082004001100004 Hasegawa S, Yoshida Y, Morimoto M, et al. Transanal TME: new standard or fad?. J Anus Rectum Colon. 2019;3(1):1-9. Published 2019 Jan 29. doi:10.23922/jarc.2018-030

s-0039-1696728

Köckerling F, von Rosen T, Jacob D. Modified plug repair with limited sphincter sparing fistulectomy in the treatment of complex anal fistulas. Front Surg. 2014;1:17. Published 2014 May 30. doi:10.3389/fsurg.2014.00017

Nuño-Guzmán CM, Hernández-Carlín JM, Almaguer FI. Colovesical, coloenteric, colocutaneous fistula, and hip

### DISCUSSION

## REFERENCES

Huda T, Pandya B. Treatment of Colocutaneous Fistula in the Left Thigh. Surg J (N Y). 2019;5(3):e113-e119. Published 2019 Sep 21. doi:10.1055/

Murphy PB, Belliveau P. Left-sided sigmoid diverticulitis presenting as right-sided thigh abscess. Int Surg. 2012;97(4):285-287. doi:10.9738/CC144.1

eptic arthritis secondary to sigmoid diverticulitis. Int J Colorectal Dis. 2010;25(6):793-794. doi:10.1007/s00384-009-0861-5