

Received: 2020.05.13

Accepted: 2020.09.16

Available online: 2020.09.28

Published: 2020.11.09

A Benign Appendiceal-Colonic Fistula, Diagnosed and Managed Laparoscopically: A Case Report

Authors' Contribution:

Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

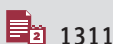
ABCDEFG

Hiroki Nakamoto
Ryoichi Yokota
Hiromasa Namba
Tomohiro Ishikawa
Kenji Yamada
Mitsuchika Hosoda
Koichi Taguchi

Department of Surgery, Sunagawa City Medical Center, Sunagawa, Hokkaido, Japan

Corresponding Author: Hiroki Nakamoto, e-mail: hiroki.nakamoto0701@gmail.com**Conflict of interest:** None declared

Patient: Male, 76-year-old
Final Diagnosis: Appendiceal-colonic fistula
Symptoms: None
Medication: —
Clinical Procedure: Appendectomy • wedge-shaped resection of rectum
Specialty: Gastroenterology and Hepatology

Objective: Unusual setting of medical care**Background:** Treatment methods for appendiceal-colonic fistulas differ greatly depending on whether lesions are benign or malignant. If the tumor is malignant, appendectomy with lymph node resection (ileoceleal resection or right hemicolectomy) should be performed. There is no consensus on the method of surgery for organs infiltrated by appendiceal cancer. Furthermore, there are no reported laparoscopic cases that could be prevented from over-surgery by laparoscopy examination or rapid intraoperative pathological examination.**Case Report:** A 76-year-old man presented with positive fecal occult blood. Lower endoscopy revealed a 10-mm tumor in the rectosigmoid colon accompanied by white moss. A biopsy showed inflammatory granulation and no malignancy. Fluorodeoxyglucose-positron emission tomography showed highly increased accumulation at the tip of the appendix, and the standardized uptake value max was 7.3. We suspected a benign lesion rather than appendiceal cancer with infiltration into the rectosigmoid colon; therefore, we performed laparoscopic appendectomy and wedge-shaped resection of the rectum of the sigmoid colon. An intraoperative rapid pathological examination showed no appearance of malignancy; therefore, additional resection was omitted, and an ileostomy was created in the right lower quadrant. A permanent pathological examination showed complicated appendicitis, with no appearance of malignancy. The ileostomy was closed on postoperative day 25, and the patient was discharged on postoperative day 32.**Conclusions:** In cases where there is difficulty in identifying whether the appendiceal-colonic fistula lesion is benign or malignant, laparoscopy and intraoperative rapid pathological examination may be useful in avoiding excessive treatment.**MeSH Keywords:** Fistula • Fluorodeoxyglucose F18 • Pathologic Processes**Abbreviations:** CEA – carcinoembryonic antigen; CA19-9 – carbohydrate antigen 19-9; FDG-PET – fluorodeoxyglucose-positron emission tomography; SUV – standardized uptake value; POD – postoperative day**Full-text PDF:** <https://www.amjcaserep.com/abstract/index/idArt/925946>

1311



—



8



15



Background

Appendiceal-colonic fistulas are rare, regardless of whether they are benign or malignant. Treatment methods for appendiceal-colonic fistulas differ greatly depending on whether lesions are benign or malignant. If the tumor is malignant, ileocecal resection or right hemicolectomy with lymph node resection should be performed. There is no consensus method of surgery for other organs infiltrated by appendiceal cancer, nor is it clear to what degree regional lymph nodes should be resected. However, metastasis has reportedly occurred in the para-intestinal lymph node through an appendiceal-colon fistula formed by appendiceal cancer. Fluorodeoxyglucose-positron emission tomography (FDG-PET) is a good preoperative option. However, as FDG-PET can be positive in inflammatory tissues, excessive treatment might be performed for benign tumors. In the present case, we admitted a patient with an appendiceal-rectosigmoid colon fistula, which was later diagnosed as a benign lesion, who was complicated by appendicitis. Excessive treatment through over-surgery could be avoided through examination laparoscopy and intraoperative rapid pathological examination.

Case Report

A 76-year-old man presented with fecal occult blood. On examination, he was 160 cm tall and weighed 55.0 kg (body mass index 20.0). There was no complaint of symptoms such as stomachache or lower abdominal pain. His white blood cell (WBC) count was slightly elevated at 10 100/ μ l and hemoglobin was 13.5g/dl; however, other hematological values were within normal ranges. Tumor markers such as carcinoembryonic antigen (CEA) and carbohydrate antigen 19-9 (CA19-9) were within normal limits. He had a history of total thyroidectomy

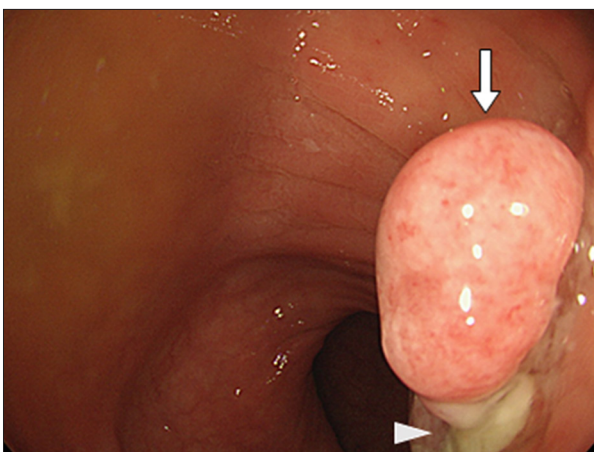


Figure 1. Lower endoscopy revealed a 10-mm tumor in the rectosigmoid colon (white arrow), surrounded by a white coat (white arrowhead).

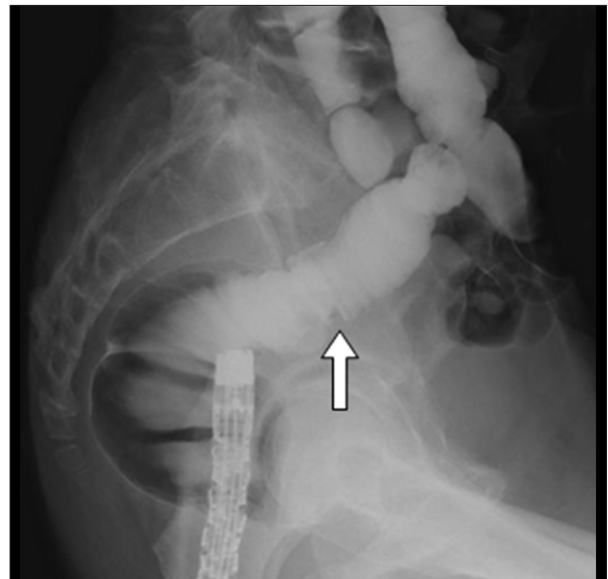


Figure 2. Enema gastrographic imaging in lateral view showed missing images of the 10-mm tumor in rectosigmoid colon (white arrow), but no appearance of a fistula or an extra-contrast agent.



Figure 3. Contrast CT showed no appearance of the tumor or swelling of the rectum lymph nodes and a huge feces stone in the appendix apex with liquid storage (white arrow).

for thyroid cancer 14 years ago and had been taking levothyroxine regularly. He also had diabetes and hyperlipidemia, and thus was on internal medications.

Lower endoscopy revealed a 10-mm tumor surrounded by a white coat in the rectosigmoid colon (Figure 1). There was no significant appearance in the opening of the appendix, and no appearance of inflammatory bowel disease or diverticulitis. Enema gastrographic imaging showed missing images of the 10-mm tumor but no appearance of a fistula or an extra-contrast

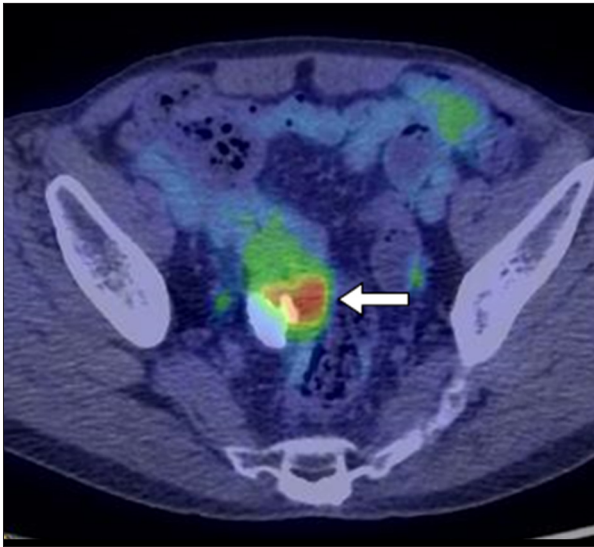


Figure 4. FDG-PET showed increased accumulation of FDG at the appendix apex; SUV max was 7.3 (white arrow).

agent (Figure 2). The biopsy of the tumor showed inflammatory granulation and no malignancy. Contrast CT showed no appearance of the tumor or swelling of the rectum lymph nodes, a fecalith at the appendix apex, or liquid storage (Figure 3). There was no appearance of diverticulitis. FDG-PET showed increased accumulation of FDG in the appendix apex, with a standardized uptake value (SUV) of 7.3 (Figure 4).

A biopsy of the tumor tissues revealed no appearance of malignancy, and showed that the rectosigmoid colon lesion was surrounded by a white coat, so we suspected a benign lesion such as complicated appendicitis rather than appendiceal cancer with infiltration into the rectosigmoid colon. A laparoscopic ileocecal resection and high anterior resection of the rectum with lymph node dissection was planned. The patient was placed in a lithotomy and head-low position. A 12-mm trocar was placed at the right lower quadrant and three 5-mm ports were placed in the left upper and lower quadrants and the right upper quadrant (Figure 5). Laparoscopic coagulation shears were used as the energy device. There was firm adhesion to the appendix apex; thus, the right-side wall of the rectum, the appendix apex, bladder, and small intestine formed a single lump. There were redness and strong edema of tissues suspecting of inflammatory changes, so we diagnosed a benign lesion such as complicated appendicitis. We resected the adhesion and the root of the appendix. There was difficulty in the resection between the appendix and the rectum, so a wedge-shaped resection of the rectosigmoid colon was performed (Figure 6). An intraoperative rapid pathological examination did not reveal any malignancy; therefore, ileocecal resection and high anterior resection of the rectum with lymph node dissection were omitted. To avoid leakage of the rectum due to increased intestinal pressure, a covering ileostomy was created in the right lower quadrant.

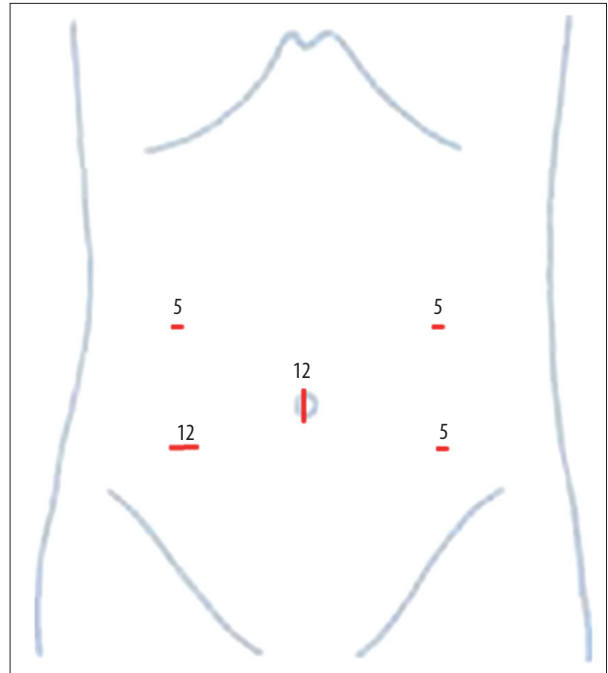


Figure 5. A 12-mm trocar was placed at the right lower quadrant and three 5-mm ports were placed in the left upper and lower quadrants and the right upper quadrant.

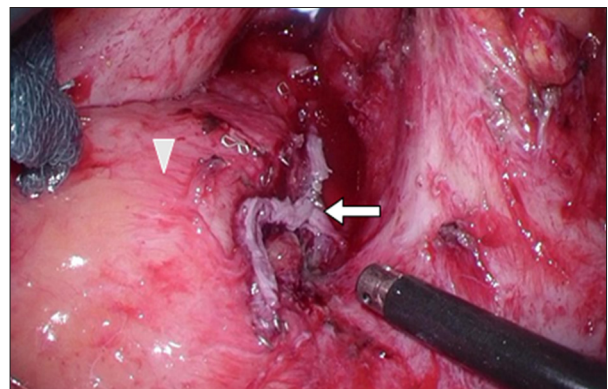


Figure 6. Laparoscopic appearance showed difficulty in resection of the adhesion between the appendix and rectum; thus, a wedge-shaped resection was conducted at the rectosigmoid colon (white arrow; resection site, white arrow head; rectosigmoid colon wall).

The operation time was 241 min and the estimated blood loss was 5 ml. There were no intraoperative complications necessitating conversion to laparotomy. The permanent pathological examination showed phlegmonous appendicitis and spread of inflammation to the deep appendix and rectal wall, with no malignant appearance (Figures 7, 8). The management of ileostomy and dehydration by drainage from ileostomy were problematic, so the ileostomy was closed on the postoperative day (POD) 25 and the patient was discharged on POD 32.

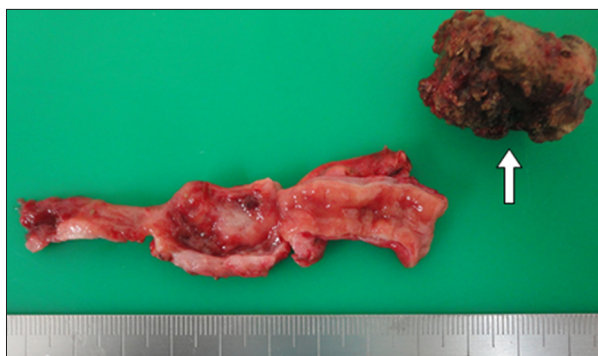


Figure 7. Extracted, opened specimens. There was a fecalith in the tip of the appendix (white arrow).

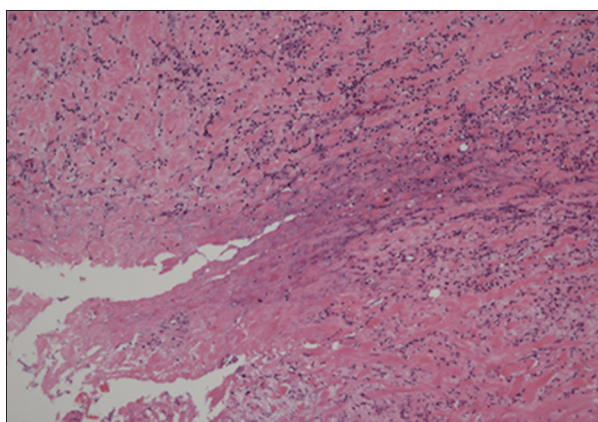


Figure 8. Permanent pathological examination showed phlegmonous appendicitis and spread of inflammation to the deep appendix and rectum wall, with no malignant appearance. (Hematoxylin and eosin staining; $\times 100$).

At a follow-up examination 3 months after closure of the ileostomy, no complications were found.

Discussion

Appendiceal-colonic fistulas are rare. The most common causes of fistulas are benign diseases, such as abscess-forming appendicitis; however, there have been cases reported in which the cause was malignant (e.g., appendiceal cancer) [1]. The presence of a fistula can be confirmed through enema gastrographic imaging [2]. In this case, we considered that an appendiceal-colonic fistula existed; however, the fistula disappeared by drainage of the abscess to the lumen of the rectum; therefore, there was no appearance of a fistula or an extra-contrast agent.

The treatment strategy for appendiceal-colonic fistulas differs greatly between benign and malignant lesions. If the tumor is benign, appendectomy and fistula resection is considered [3].

However, if the tumor is malignant, appendectomy with lymph node resection (ileocecal resection or right hemicolectomy) should be considered [4–7]. There is no consensus on the method of surgery for organs infiltrated by appendiceal cancer or the degree to which regional lymph nodes should be resected. However, there is a case report in which metastasis to the para-intestinal #251 lymph node in an appendiceal-rectosigmoid colon fistula occurred through appendiceal cancer [8]; therefore, metastasis of appendiceal cancer to lymph nodes of the rectum is possible. If there is suspicion of appendiceal cancer and invasion to the surrounding colon, ileocecal resection, right hemicolectomy, and infiltrated colon resection with lymph node resection should be considered. Thus, the treatment strategies for appendiceal-colonic fistulas differ greatly for benign vs. malignant lesions.

The FDG-PET testing is useful in discriminating between benign or malignant tumors [9]. However, benign lesion can be positive due to increased accumulation of FDG, such as physiological accumulation of brain or urinary tract, inflammatory lesion, abscess, and traumatic changes [10,11]; therefore, it must be considered that a positive FDG-PET result might be a false positive. Essentially, when we cannot completely diagnose the tumor as either benign or malignant, we favor malignancy and remove the tumor completely; however, this might lead to excessive treatment of benign tumors. In 2 past cases, appendiceal cancer was diagnosed because SUV max was 7.3 and 17.1, respectively. Ileocecal resection with lymph node resection were performed; however, there was no malignant appearance in the permanent pathological examination [12]. SUV max does not always reflect the degree of malignant disease. Therefore, various methods to elevate the sensitivity and specificity of FDG-PET have been reported. Studies to reduce the possibility of false positives have been conducted using labeled amino acid PET scans instead of FDG scans [13], and in a comparison between FDG and 3-deoxy-3-¹⁸F-fluorothymidine (FLT) in rats, the tumor specificity of FLT was reported to be higher than that of FDG [14]. ¹⁸F-fluoroethyl-L-tyrosine has been reported to have greater ability to discriminate between tumors and inflammation and to have higher specificity for tumor diagnosis than that of FDG [15]. These studies may suggest a useful method for improving the discrimination between benign vs. malignant tumors.

Intraoperative rapid pathological diagnosis can be a good option for avoiding excessive treatment. The present case is the first to be reported in which over-surgery could have been prevented by laparoscopy and rapid intraoperative pathological examination. However, there may be cases in which the rapid pathological diagnosis differs from the permanent diagnosis; thus, in place of using intraoperative rapid pathological diagnosis, sufficient informed consent before surgery is needed.

Conclusions

In cases where it is difficult to identify whether an appendiceal-colonic fistula lesion is benign or malignant, laparoscopic and intraoperative rapid pathological examination may be useful in avoiding excessive treatment.

References:

1. Suwa H, Baba N, Demura K et al: A case of appendiceal -- sigmoid fistula. *Jpn J Gastroenterol Surg*, 2002; 35: 194–98
2. Morris-Stiff GJ, Islam KA: Appendico-colic fistula complicating appendicitis in cystic fibrosis. *BMJ Case Rep*, 2010; 2010: bcr0220102714
3. Joo JS, Agachan F, Wexner SD: Laparoscopic surgery for lower gastrointestinal fistulas. *Surg Endosc*, 1997; 11: 116–18
4. Tang LH, Shia J, Soslow RA et al: Pathologic classification and clinical behavior of the spectrum of goblet cell carcinoid tumors of the appendix. *Am J Surg Pathol*, 2008; 32: 1429–43
5. Knigge U, Hansen CP: [Appendical carcinoid tumors and goblet cell carcinoids.] *Ugeskr Leager*, 2010; 172: 1678–81 [in Danish]
6. Pham TH, Wolff B, Abraham SC, Drelichman E: Surgical and chemotherapy treatment outcomes of goblet cell carcinoid: A tertiary cancer center experience. *Ann Surg Oncol*, 2006; 13: 370–76
7. Varisco B, McAlvin B, Dias J, Franga D: Adenocarcinoid of the appendix: Is right hemicolectomy necessary? A meta-analysis of retrospective chart reviews. *Am Surg*, 2004; 70: 593–99
8. Toyozumi T, Ohira M, Miyauchi H et al: A case of appendiceal carcinoma with direct invasion to the ascending colon. *Chiba Medical Journal*, 2015; 91: 113–17
9. Mori T, Mizuno R, Itou D et al: Useful FDG-PET for preoperative diagnosis of primary carcinoma of the vermiform appendix – a case report. *J Jpn Surg Assoc*, 2009; 70: 778–82
10. Rosenbaum SJ, Lind T, Antoch G et al: False-positive FDG-PET uptake – the role of PET/CT. *Eur Radiol*, 2005; 17: 1–12
11. Klaff V, Rodney JH, Robert EW et al: The clinical impact of 18F-FDG PET in patients with suspected or confirmed recurrence of colorectal cancer: A prospective study. *J Nucl Med*, 2002; 43: 492–99
12. Ogawa S, Itabashi M, Kameoka S: Significance of FDG-PET in identification of diseases of the appendix – based on experience of two cases falsely positive for FDG accumulation. *Case Rep Gastroenterol*, 2009; 29: 125–30
13. Lim JW, Tang CL, Keng GH: False-positive F-18 fluorodeoxyglucose combined PET/CT scans from suture granuloma and chronic inflammation: Report of two cases and review of literature. *Ann Acad Med Singapore*, 2005; 34: 457–60
14. Van Waarde A, Cobben DC, Suurmeijer AJ et al: Selectivity of 18F-FLT and 18F-FDG for differentiating tumor from inflammation in a rodent model. *J Nucl Med*, 2004; 45: 695–700
15. Kaim AH, Weber B, Kurrer MO et al: 18F-FDG and 18F-FET uptake in experimental soft tissue infection. *Eur J Nucl Med*, 2002; 29: 648–54

Acknowledgements

We thank all staff members at the Sunagawa City Medical Center.

Conflict of interests

None.