

Journal of Surgical Case Reports, 2021;3, 1-4

doi: 10.1093/jscr/rjab086 Case Report

CASE REPORT

Potential problems of partial resection for colitis-associated cancer in a patient with ulcerative colitis: case report

Yuki Horio^{1,*}, Motoi Uchino¹, Takako Kihara², Toshihiro Bando¹, Ryuichi Kuwahara¹, Tomohiro Minagawa¹, Kurando Kusunoki¹, Kenji Watanabe³, Seiichi Hirota² and Hiroki Ikeuchi¹

¹Department of Inflammatory Bowel Disease Surgery, Hyogo College of Medicine, Hyogo, Japan, ²Department of Surgical Pathology, Hyogo College of Medicine, Hyogo, Japan and ³Division of Internal Medicine, Department of Inflammatory Bowel Disease, Hyogo College of Medicine, Hyogo, Japan

*Correspondence address. Department of Inflammatory Bowel Disease, Hyogo College of Medicine, 1-1 Mukogawa-cho, Nishinomiya, Hyogo 663-8501, Japan. Tel: +81-798-45-6372; Fax: +81-798-45-6373; E-mail: yu-horio@hyo-med.ac.jp

Abstract

Total proctocolectomy and an ileal pouch-anal anastomosis are recommended as the standard procedure for ulcerative colitis (UC)-colitis-associated cancer (CAC). However, several studies have reported the partial colectomy and endoscopic resection of UC-CAC in recent years. We present a surgical case of UC-CAC that was detected at a site that had not been diagnosed preoperatively, and we report potential problems of partial colectomy and endoscopic resection through this case. Considerations of synchronous and metachronous cancer/dysplasia are important before partial resection is planned for CAC in UC. Moreover, it should be noted that endoscopic resection at the anal site can be a risk factor for pouch surgery failure due to fibrosis after resection.

INTRODUCTION

Ulcerative colitis (UC) is steadily increasing in Japan due to the aging of the population and the increase in the number of longterm patients caused by the progress of medical treatment. The number of patients who require surgery for colitis-associated cancer (CAC) is also increasing [1]. Total proctocolectomy and an ileal pouch-anal anastomosis (IPAA) are recommended as the standard procedure for UC-CAC/high-grade dysplasia [2]. Conversely, several studies have reported the partial resection and curative endoscopic submucosal dissection (ESD) of neoplasms in UC, and surveillance colonoscopy is recommended after complete removal via endoscopic resection [3, 4]. However, there are still many problems, such as the lack of long-term outcomes and the difficulty of the procedure.

We report a case of UC-CAC that was detected at a site that had not been diagnosed preoperatively.

CASE

A 56-year-old man was diagnosed with UC at the age of 30. He was controlled with 5-aminosalicylic acid and enemas and

Received: February 4, 2021. Accepted: February 16, 2021

Published by Oxford University Press and JSCR Publishing Ltd. All rights reserved. © The Author(s) 2021.

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (http://creativecommons.org/ licenses/by-nc/4.0/), which permits non-commercial re-use, distribution, and reproduction in any medium, provided the original work is properly cited. For commercial re-use, please contact journals.permissions@oup.com



Figure 1: (A) Endoscopic findings indicated Isp-type lesions in the lower rectum (arrowhead). (B) It was difficult to secure adequate margins by snaring during endoscopic mucosal resection due to contact with the dentate line. (C and D) There were no findings of cancer in ascending colon.

was treated with prednisolone at the time of exacerbation. He underwent regular surveillance endoscopy. At the age of 56, the patient was in remission. An Isp-type lesion was detected on the lower rectum, and it was diagnosed as group 5. Endoscopic findings are shown in Fig. 1. Isp-type lesions were found in the lower rectum near the dentate line. There were no findings of cancer in other sites of the pan-colon by surveillance endoscopy. Endoscopic mucosal resection (EMR) was tried to distinguish between sporadic cancer and CAC. Although the negative nonlifting sign was confirmed, EMR could not be performed due to the difficulty of snaring. Finally, the patient was recommended to choose ESD or surgery. Therefore, he was referred to our department for surgery at his own will.

The physical findings on admission were as follows: blood pressure 129/70 mmHg, pulse 72 times/min, body temperature 37.3°, stool frequency 1–2 times/day and no bloody stools. The blood biochemical test findings indicated a hemoglobin level of 13.1 g/dl and an erythrocyte sedimentation rate of 5 mm (1 H), which was normal. The UC severity was mild by using the criteria of Truelove and Witts [5]. The disease extent of UC was pancolitis. Tumor markers were normal: carcinoembryonic antigen: 1.1 ng/ml and carbohydrate antigen 19-9: 10.4 U/ml. By computed tomography, tumors could not be identified, and multiple diverticula were found in the ascending colon, descending colon and sigmoid colon. No other obvious abnormalities were observed, and no metastatic lesions were found.

The patient underwent laparoscopic-assisted total proctocolectomy, mucosectomy, handsewn IPAA and ileostomy



Figure 2: The resected specimen shows a 15 \times 14 mm Ip lesion that was found in the ascending colon (arrow head), and a 5 mm-sized elevated lesion was observed in the lower rectum (dotted line circle): it is difficult to recognize with the naked eye.

construction. The surgical findings were no ascites and no obvious metastatic lesions. The resected specimen is shown in Fig. 2.

The histopathological findings were as follows: a 15×14 mm Ip lesion was found in the ascending colon, and a welldifferentiated tubular adenocarcinoma showing moderate atypia was found in the tubular lineage. In the lower rectum,



Figure 3: (A-1) Histopathological findings. A 15×14 mm Ip lesion was found in the ascending colon, and a well-differentiated tubular adenocarcinoma showing moderate atypia was found in the tubular lineage (arrow). (A-2) Ki-67 was highly positive, whereas p53 was strongly positive in a diffuse manner. (A-3) p53 was weakly positive in a mosaic pattern and in the cancerous component. (B) In the lower rectum, a 5 mm-sized elevated lesion was observed, and a low-grade dysplasia-equivalent image showing moderate atypia was also obtained (arrowhead). The resected margin, which was 11 mm in size, was negative.

a 5 mm-sized elevated lesion was observed, and a low-grade dysplasia-equivalent image showing moderate atypia was also obtained. The resected margin, which was 11 mm in size, was negative. No lymph node metastasis was observed (Fig. 3). The 8th Union for International Cancer Control Pathological TNM staging system classification was A, 0-Ip, tub1, pTis (M), ly0, v0, pN0 (0/11), pPM0, pDM0 and stage 0.

DISCUSSION

Total proctocolectomy is recommended as the standard procedure for UC-CAC/high-grade dysplasia in the Japan guidelines, ECCO guidelines and practice parameters of the American Society of Colon and Rectal Surgeons [2, 6, 7]. Partial resection and ileorectal anastomosis have limited indications [8, 9]. The reasons include the risks of a high frequency of multiple cancer lesions and the development of CAC in the remnant colon [10]. Regarding synchronous cancer, Hata et al. [11] reported that 17 out of 238 (7.14%) cases with invasive CAC were unrecognized preoperatively. For these cases, it was not enough just to perform a partial resection, and they required reoperation immediately. Regarding metachronous cancer, there is no evidence of long-term outcomes for metachronous UC-CAC after segmental resection. Even if we detect synchronous cancer reliably and we perform a partial resection, there is no guarantee that metachronous cancer can be detected after surgery. In addition, the kind of surgery that can be performed in the reoperation becomes a further problem. The most common sites of UC-CAC are the rectum and sigmoid colon, and therefore, the performance of several repeated pelvic surgeries would probably

make future pouch surgery difficult owing to adhesion or pelvic infection.

Regarding endoscopic treatment for dysplasia, the SCENIC statement of the international consensus meeting held in 2014 stated, 'After complete removal of endoscopically resectable dysplastic lesions, surveillance colonoscopy is recommended rather than colectomy' [12]. However, there are many problems, such as that the definition of endoscopically resectable lesions has not been determined, the procedure is difficult due to submucosal fibrosis and the long-term outcome is still unclear. With this assumption, accurate diagnosis is a prerequisite for performing endoscopic treatment. In the present case, ascending colon cancer that was not diagnosed before surgery was detected during the pathology analysis of an excised colorectal specimen. In addition, if there is anal transition zone cancer, endoscopic diagnosis and treatment will be difficult. Even if either EMR or ESD was performed, these procedures lead to surgical anal canal fibrosis and stenosis, and surgeons may encounter difficulties with mucosectomy and insertion of the circular stapler.

Although there are several reports recommending partial recession, EMR and ESD, these procedures should not be taken for granted, as there is still low-grade evidence. In this case report, the physicians had to determine the treatment strategies considering various issues such as unrecognized synchronous cancer, metachronous cancer and future surgical plans. We summarized these potential problems in a schema (Fig. 4).

Considerations about synchronous and metachronous cancer/dysplasia are important before the planning of partial resection for CAC in UC. Moreover, it should be mentioned that



Figure 4: Potential problems of segmental colectomy and endoscopic treatment: • Metachronous cancer after ileocecal resection: small intestine and mesentery adhesions to the right retroperitoneum that make creating the pouch and reaching the anus difficult.

Metachronous cancer after low anterior resection makes future pouch surgery
difficult owing to adhesion or pelvic infection.

Postendoscopic mucosal resection (EMR) or endoscopic submucosal dissection (ESD) in the surgical anal canal.

These procedures lead to surgical anal canal fibrosis and stenosis, and surgeons may encounter difficulties with mucosectomy and insertion of the circular stapler.

endoscopic resection at the anal site can be a risk factor for failure of IPAA due to fibrosis after resection.

AUTHORS' CONTRIBUTIONS

Y.H. drafted the article. Y.H., T.B., R.K., T.M. and K.K. treated and observed the patient. K.H. and S.H. carried out the histopathological diagnosis. M.U., K.W. and H.I. contributed towards drafting the article or revising it critically for important intellectual content and providing final approval. All authors have read and approved the manuscript.

CONSENT

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

CONFLICT OF INTEREST

None declared.

FUNDING

None declared.

ABBREVIATIONS

IPAA, ileal pouch-anal anastomosis; UC, ulcerative colitis; CAC, colitis-associated cancer; EMR, endoscopic mucosal resection; ESD, endoscopic submucosal dissection.

REFERENCES

- Uchino M, Ikeuchi H, Hata K, Okada S, Ishihara S, Morimoto K, et al. Changes in the rate of and trends in colectomy for ulcerative colitis during the era of biologics and calcineurin inhibitors based on a Japanese nationwide cohort study. Surg Today 2019;49:1066–73.
- Magro F, Gionchetti P, Eliakim R, Ardizzone S, Armuzzi A, Barreiro-de Acosta M, et al. Third European evidencebased consensus on diagnosis and management of ulcerative colitis. Part 1: definitions, diagnosis, extra-intestinal manifestations, pregnancy, cancer surveillance, surgery, and ileo-anal pouch disorders. J Crohns Colitis 2017;11: 649–70.
- Iacopini F, Saito Y, Yamada M, Grossi C, Rigato P, Costamagna G, et al. Curative endoscopic submucosal dissection of large nonpolypoid superficial neoplasms in ulcerative colitis (with videos). Gastrointest Endosc 2015;82: 734–8.
- 4. Frontali A, Cohen L, Bridoux V, Myrelid P, Sica G, Poggioli G, et al. Segmental colectomy for ulcerative colitis: is there a place in selected patients without active colitis? An international multicentric retrospective study in 72 patients. *J Crohns* Colitis 2020;**14**:1687–92.
- 5. Truelove SC, Witts LJ. Cortisone in ulcerative colitis; final report on a therapeutic trial. Br Med J 1955;2:1041–8.
- 6. Matsuoka K, Kobayashi T, Ueno F, Matsui T, Hirai F, Inoue N, et al. Evidence-based clinical practice guidelines for inflammatory bowel disease. *J Gastroenterol* 2018;**53**:305–53.
- 7. Ross H, Steele SR, Varma M, Dykes S, Cima R, Buie WD, et al. Practice parameters for the surgical treatment of ulcerative colitis. Dis Colon Rectum 2014;**57**:5–22.
- Lightner AL, Tse CS, Quinn K, Bergquist JR, Enders F, Pendegraft R, et al. Preoperative Clostridium difficile infection does not affect pouch outcomes in patients with ulcerative colitis who undergo ileal pouch-anal anastomosis. *Inflamm Bowel* Dis 2017;23:1195–201.
- 9. Uzzan M, Kirchgesner J, Oubaya N, Amiot A, Gornet JM, Seksik P, et al. Risk of rectal neoplasia after colectomy and ileorectal anastomosis for ulcerative colitis. J Crohns Colitis 2017;11:930–5.
- Watanabe T, Konishi T, Kishimoto J, Kotake K, Muto T, Sugihara K. Ulcerative colitis-associated colorectal cancer shows a poorer survival than sporadic colorectal cancer: a nationwide Japanese study. *Inflamm Bowel Dis* 2011;17:802–8.
- Hata K, Anzai H, Ikeuchi H, Futami K, Fukushima K, Sugita A, et al. Surveillance colonoscopy for ulcerative colitisassociated colorectal cancer offers better overall survival in real-world surgically resected cases. Am J Gastroenterol 2019;114:483–9.
- Laine L, Kaltenbach T, Barkun A, McQuaid KR, Subramanian V, Soetikno R, et al. SCENIC international consensus statement on surveillance and management of dysplasia in inflammatory bowel disease. *Gastrointest Endosc* 2015;81: 489–501.e26.