

[CASE REPORT]

Double Early Rectal Cancer Arising from Multiple Inflammatory Cloacogenic Polyps Resected by Endoscopic Submucosal Dissection

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Abstract:

A 45-year-old man visited our institution due to the onset of hematochezia. He had a previous episode nine years earlier and colonoscopy at that time revealed multiple polyps, which were consistent with inflammatory cloacogenic polyps (ICPs) on the dentate line. Colonoscopy was performed again and two of the ICPs had grown. Both lesions were pathologically diagnosed as adenocarcinomas based on biopsies. Endoscopic submucosal dissection (ESD) was performed and the two lesions were diagnosed as double well-differentiated adenocarcinomas arising from ICPs. To our knowledge, this is the first reported case of double early rectal cancer in ICPs, which were followed endoscopically and successfully resected with ESD.

Key words: inflammatory cloacogenic polyp, mucosal prolapse syndrome, solitary rectal ulcer syndrome, rectal cancer, endoscopic submucosal dissection

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Introduction

An inflammatory polyp is a concept that broadly refers to a mucosal bulge associated with inflammation and the term is not yet clearly defined. Inflammatory cloacogenic polyp (ICP) is a type of inflammatory polyp which is typically a prominent lesion arising from the transitional epithelium of the anal canal. It is characterized by a tubulovillous pattern of growth and superficial ulceration. Lobert et al. first described this lesion in 1981 (1). The concept is also included in the raised type of rectal mucosal prolapse syndrome (2). Inflammatory polyps are said to be benign, and there have been few reports of cancer arising from them to date. Moreover, there are no reports on ICPs with malignancy that were resected with endoscopic submucosal dissection (ESD). We herein report on a rare case of double rectal cancer arising from ICPs which were successfully treated by ESD.

Case Report

A 45-year-old man visited our institution due to the onset of hematochezia. His family history included colorectal cancer in his father. He had a previous episode nine years earlier and colonoscopy at that time revealed multiple polyps, which were consistent with ICPs on the dentate line (Fig. 1A, B). Biopsies from these polyps at that time were diagnosed as inflammatory polyps. Colonoscopy at this presentation revealed multiple polyps in the lower rectum as before, two of which had grown in size, and the shapes of the top were different from surrounding polyps (Fig. 1C, D). Biopsies were performed for the two polyps and the pathological diagnoses were adenocarcinomas for both lesions. Additional endoscopy revealed abnormalities in both microvascular and microsurface patterns in narrow band imaging (NBI) magnifying endoscopy (Fig. 2A, C), which consisted with the Japan NBI Expert Team (JNET) classification type 2 B (3) and type V_I-mildly irregular pit pattern (4) in magnifying chromoendoscopy with crystal violet staining

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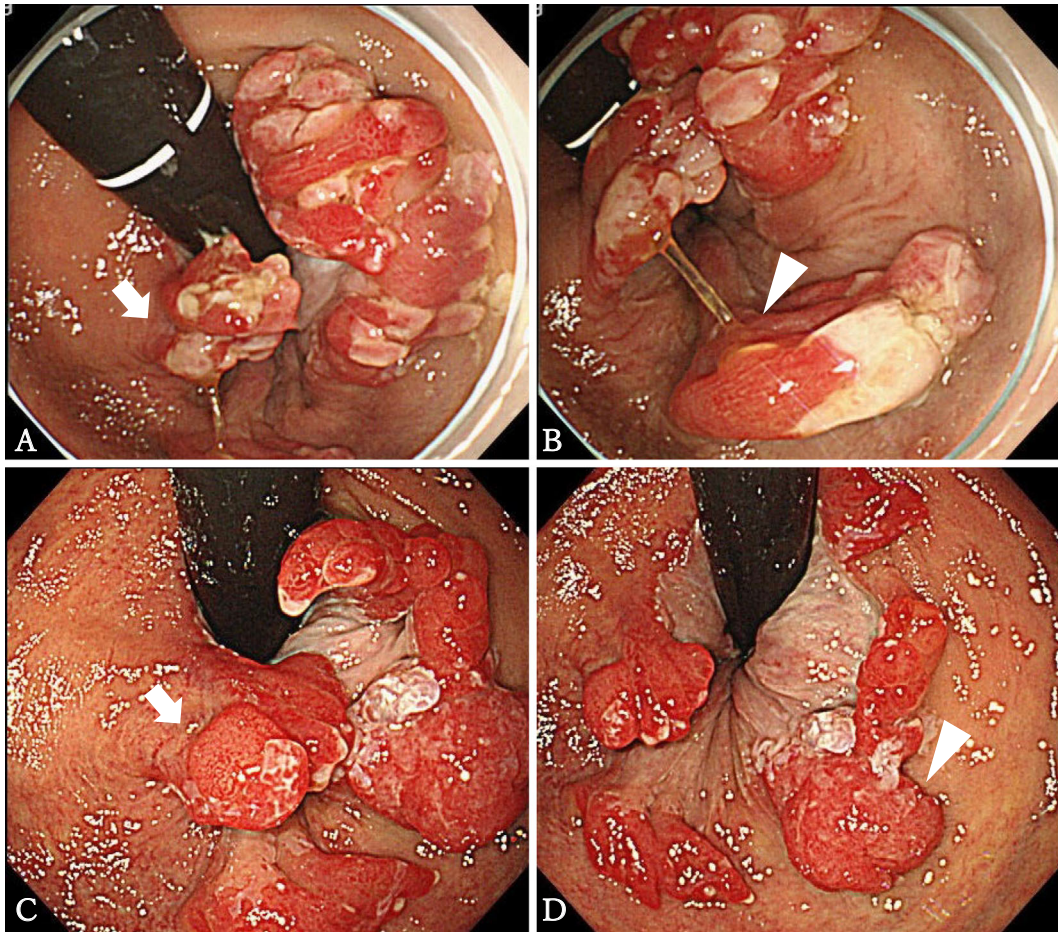


Figure 1. White light endoscopic findings of the lesions 9 years earlier (A, B) and before endoscopic submucosal dissection (C, D). White arrow shows lesion No.① and white triangle shows lesion No.②.

(Fig. 2B, D), which were consistent with cancer. In contrast, the base of each polyp and other polyps were consistent with inflammatory polyp (Fig. 2E, F). Therefore, we performed ESD using a dual knife (Olympus Co., Ltd., Tokyo, Japan). The two polyps, diagnosed as adenocarcinoma, were resected separately en bloc with nearby ICPs (Fig. 2G). During the procedure, there were no findings such as fibrosis or non-lifting sign, however, thick vessels in the submucosa were observed. Additionally, the remaining ICPs were removed with endoscopic mucosal resection (EMR). After ESD, fever and small amount of bleeding during defecation were observed for two and seven days respectively, however, both gradually improved. In the two polyps resected by ESD, the pathological diagnosis was early rectal cancer as follows: ① 8×6 mm, well-differentiated adenocarcinoma (tub1), Tis, Ly0, V0, margin negative; and ② 16×13 mm, tub1, pTis, Ly0, V0, margin negative (Fig. 3, 4). The other two polyps subjected to EMR were inflammatory polyps. These multiple polyps showed similar pathological features, such as smooth muscle proliferation into the lamina propria, mild fibrosis of the stroma, and hyperplastic glandular epithelium. Endoscopic and pathological findings suggested that the two rectal cancer had arisen from the ICPs. After

discharge, the patient had no hematochezia and follow-up colonoscopy performed 10 months after ESD showed no recurrence in the rectum (Fig. 2H).

Discussion

Inflammatory polyps are a type of non-neoplastic colorectal polyps classified into several types according to shape, background, and pathological analysis. In our case, there was no background of inflammatory bowel disease, and no polyps were found except at the dentate line. The inflammatory polyps were localized on the dentate line and the pathological diagnosis was thus consistent with ICPs, a component of rectal mucosal prolapse syndrome (MPS) first reported as solitary rectal ulcer syndrome in 1969 (5). Our patient had a habit of straining at defecation, therefore, the diagnosis of ICPs based on MPS is plausible.

There have been few reports on inflammatory polyps associated with cancer. Table shows the reported cases of ICPs associated with neoplasm. Parfitt et al. described pathological findings of ICPs complicating rectal adenomas and adenocarcinoma (6). In addition, Hanson et al. reported on a case with anal intraepithelial neoplasia in an ICP (7) and Ja-

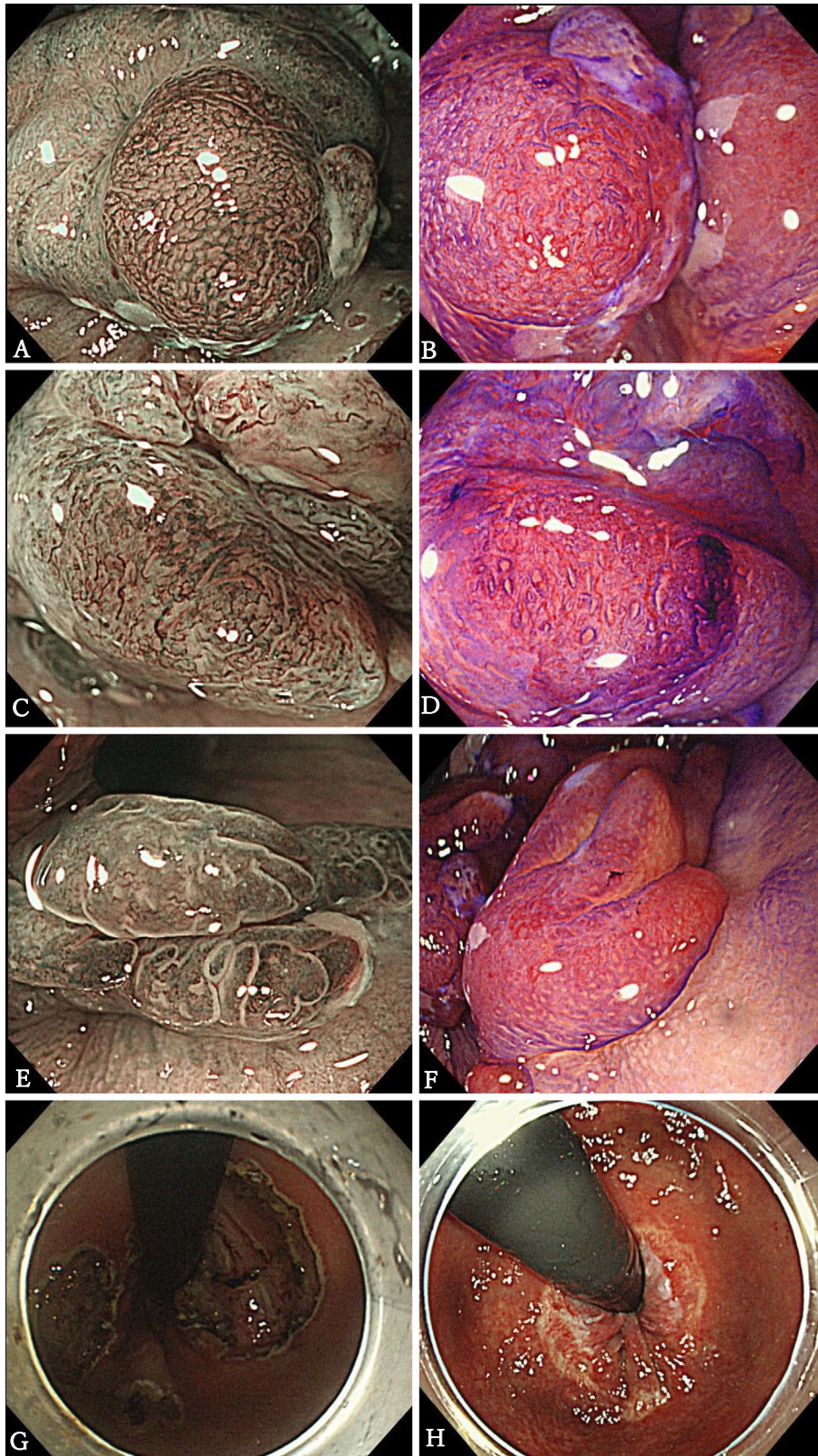


Figure 2. Narrow band imaging endoscopy findings, crystal violet staining, and comparison before and after endoscopic submucosal dissection (ESD). A and B show lesion No.①. C and D show lesion No.②. E and F show an inflammatory cloacogenic polyp. G shows ESD ulcer at the time of the procedure; H shows an ESD scar 10 months after the procedure.

worski et al. described squamous cell carcinoma arising in ICPs (8). In both reports, human papillomavirus was considered to be associated with these neoplasms. Additionally,

few studies have so far reported on patients with MPS accompanied by adenocarcinoma (9). Although there has been speculation about malignant transformation from MPS (10),

this remains controversial and unclear.

Of note, our patient had undergone endoscopy nine years before this presentation, so we could follow the endoscopic and pathologic changes of the ICPs; this is in contrast to almost all other reported cases, which had not been followed up endoscopically. At previous endoscopy, the polyps were relatively smaller and the pathological diagnoses at the time were benign inflammatory polyps. In contrast, nine years later, the size and shape of the lesions had changed and pathological diagnosis from biopsy of these two lesions was adenocarcinoma. It would be difficult to determine whether the cancer arose from the ICPs, however, we believe that our endoscopic observation and pathological results could point to carcinogenesis arising from ICPs.

In terms of the treatment of such lesions, only one report describes ESD performed for rectal cancer associated with MPS (11). In our case, ESD was performed for double early

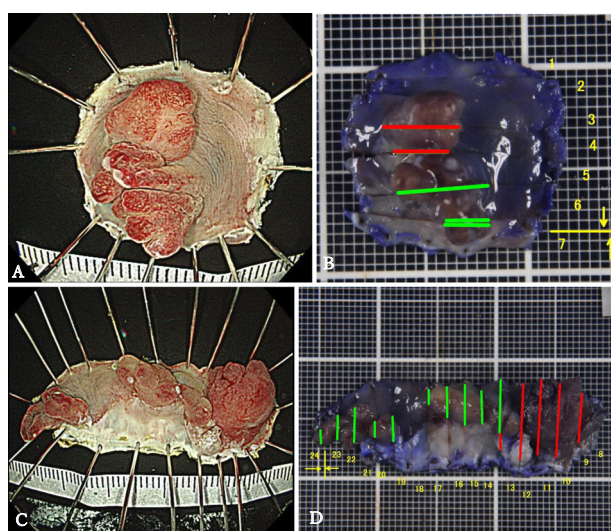


Figure 3. Specimens resected by endoscopic submucosal dissection (ESD) (A and B; lesion No. ①. C and D; lesion No. ②). Red line in image B and D shows the areas of adenocarcinoma. Green line in image B and D shows the areas of inflammatory polyp.

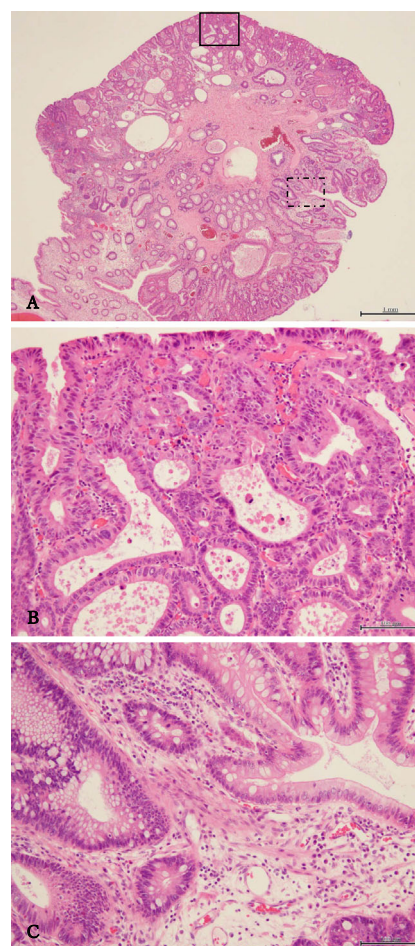


Figure 4. Histological examination of lesion No. ① (slice no. 3). (A) Hematoxylin and Eosin (H&E) staining shows intramucosal adenocarcinoma in inflammatory cloacogenic polyp (ICP). (B) Area of intramucosal adenocarcinoma framed by solid box in A (H&E staining), showing irregular, fused, and back-to-back cribriform glands. (C) Area framed by dotted box in image A (H&E staining) showing hyperplastic crypt epithelium, mild architectural distortion of the crypts, smooth muscle proliferation in the lamina propria, and mild inflammation, consistent with ICP.

Table. Clinicopathological Characteristics of Inflammatory Cloacogenic Polyps Associated with Neoplasm.

No.	Reference	Age (year)	Sex	Symptoms	Treatments	Pathological findings
1	6	36	M	Not described	ER	Adenoma
2	6	55	M	Not described	ER	Adenoma
3	6	62	F	Not described	ER	Adenocarcinoma (Tis)
4	6	28	F	Not described	ER	Adenoma
5	7	34	F	Rectal bleeding	ER	Moderate and severe squamous intraepithelial neoplasia associated with HPV
6	8	38	F	Rectal bleeding	ER	Squamous cell carcinoma in situ associated with HPV
7	8	41	F	Rectal bleeding	Not described	Squamous cell carcinoma in situ associated with HPV
8	Our case	45	M	Hematochezia	ER (ESD)	Double adenocarcinomas (Tis)

M: male, F: female, ER: endoscopic resection, ESD: endoscopic submucosal dissection, HPV: human papillomavirus

rectal cancer found at the top of ICPs. It can thus be said that this is a rare case because double adenocarcinomas were detected at the top of ICPs simultaneously, for which en bloc resection was performed using ESD. After the treatment, the present case had no hematochezia and follow-up colonoscopy showed no recurrence of cancer and ICPs in the rectum. A previous study from Japan described two cases with raised type of MPS, which were successfully treated by ESD (12). Both patients had bleeding during defecation, however it improved after being treated. Based on these findings, endoscopic resection might be useful not only for early cancer but also a raised type of MPS. The additional accumulation of similar cases is required.

In conclusion, we herein described a rare case of double early rectal cancer, followed up endoscopically and successfully treated with ESD.

The authors state that they have no Conflict of Interest (COI).

References

1. Lobert PF, Appelman HD. Inflammatory cloacogenic polyp. A unique inflammatory lesion of the anal transitional zone. *Am J Surg Pathol* **5**: 761-766, 1981.
2. Saul SH. Inflammatory cloacogenic polyp: relationship to solitary rectal ulcer syndrome/mucosal prolapse and other bowel disorders. *Hum Pathol* **18**: 1120-1125, 1987.
3. Sano Y, Tanaka S, Kudo SE, et al. Narrow-band imaging (NBI) magnifying endoscopic classification of colorectal tumors proposed by the Japan NBI Expert Team. *Dig Endosc* **28**: 526-533, 2016.
4. Kanao H, Tanaka S, Oka S, et al. Clinical significance of type V (I) pit pattern subclassification in determining the depth of invasion of colorectal neoplasms. *World J Gastroenterol* **14**: 211-217, 2008.
5. Madigan MR, Morson BC. Solitary ulcer of the rectum. *Gut* **10**: 871-881, 1969.
6. Parfitt JR, Shepherd NA. Polypoid mucosal prolapse complicating low rectal adenomas: beware the inflammatory cloacogenic polyp! *Histopathology* **53**: 91-96, 2008.
7. Hanson IM, Armstrong GR. Anal intraepithelial neoplasia in an inflammatory cloacogenic polyp. *J Clin Pathol* **52**: 393-394, 1999.
8. Jaworski RC, Biankin SA, Baird PJ. Squamous cell carcinoma in situ arising in inflammatory cloacogenic polyps: report of two cases with PCR analysis for HPV DNA. *Pathology* **33**: 312-314, 2001.
9. Nonaka T, Inamori M, Kessoku T, et al. A case of rectal cancer arising from long-standing prolapsed mucosa of the rectum. *Intern Med* **50**: 2569-2573, 2011.
10. Tsuchida K, Okayama N, Miyata M, et al. Solitary rectal ulcer syndrome accompanied by submucosal invasive carcinoma. *Am J Gastroenterol* **93**: 2235-2238, 1998.
11. Honda H, Nakamura K, Ishii N, Suzuki K, Fukuda K. Endoscopic submucosal dissection of rectal cancer close to the dentate line accompanied by mucosal prolapse syndrome. *ACG Case Rep J* **5**: e 9, 2018.
12. Tsuji T, Ootani S, Aoki H, et al. Two cases of the rectal mucosal prolapse syndrome in which endoscopic submucosal dissection was effective. *Gastroenterol Endosc* **55**: 250-256, 2013 (in Japanese, Abstract in English).

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