Solitary Ulcer Syndrome of the Rectum in an Aged Patient Treated With Sigmoidostomy

Masakazu Ohno[1], Takeshi Nakamura[1], Yoshiki Tabuchi[2], and Yosio Nogi[3]

A rare patient with solitary ulcer syndrome of the rectum was reported. A 78-year-old male with a habit of strong straining on defecation noticed anal bleeding of 2 weeks' duration and consulted us. Preoperative examinations revealed one bleeding ulcer with raised margin at the anterior wall of the rectum. The ulcer was pathologically diagnosed as solitary ulcer syndrome of the rectum from its characteristic findings of the biopsy-specimens. Sigmoidostomy was done, because the conservative treatment had no effect on the symptoms and signs and the patient remained in a poor general condition. Since the bleeding stopped completely 2 weeks after surgery and the ulcer became scar phase, sigmoidostoma was closed 4 weeks later. Thereafter, the recurrence was not found 34 months after operation. The pathogenesis as well as surgical treatments for this rare syndrome are discussed, because the considerable uncertainty has yet remained.

Key Words

Solitary rectal ulcer syndrome,
Surgical treatment,
Sigmoidostomy.

Introduction

Solitary ulcer of the rectum is a rare chronic rectal ulceration reported in the first comprehensive review by Madigan and Morson[1]. Although the characteristics of the symptoms are anal bleeding and pain often with straining due to long history of defecation difficulty[1, 2], the pathogenesis has yet been unclear. The lesions have been reported to show not always ulcerated but also raised ones, and multiple ulcers have been found in some cases[1-4]. In order to account for this complex disease, the term "solitary ulcer syndrome of the rectum" has been proposed by Rutter and Riddel in 1975[2]. Thereafter, this term has gained wide acceptance.

Recently, we encountered an aged male with solitary ulcer syndrome of the rectum who was treated with sigmoidostomy, because of the continuous bleeding even by the conservative treatment. Thus, the case was herein reported, and the pathogenesis and surgical treatments for this syndrome were also discussed, because the pathogenesis has yet been unclear and the surgical treatments for this syndrome have not yet been established.

Case Report

T.T., 78-year-old male with a habit of strong straining on defecation, noticed anal pain and bleeding of 2 weeks' duration and consulted us on April 17, 1996. He admitted to the Department of Sur-

First Department of Surgery[1], Faculty of Health Sciences[2], Kobe University School of Medicine, Kobe, and Department of Surgery[3], Nogi Hospital, Akashi, Japan

gery, Nogi Hospital (Akashi, Japan) 1 day later. On admission, he showed tachycardia of 103/min., but the other vital signs were within normal limits. Although the laboratory data revealed a severe inflammation and diabetes mellitus with leukocyte count of 22,600/mm^3, C-reactive protein of 13.5 mg/dl and blood sugar of 532 mg/ml, fever was not found. Anal bleeding continued after admission even by the conservative managements consisting of a high roughage diet, insulin treatment and laxatives together with avoidance of straining on defecation, and the blood examination 10 days after admission showed anemia with a hemoglobin of 7.1 g/dl and hematocrit of 21.5%.

External hemorrhoids without bleeding were found by inspection and anoscopy. A rectal ulcer at the anterior rectal wall was also found by digital examination. Barium enema showed a deep ulcer at the anterior wall of the rectum about 4 cm from anal verge (Fig. 1). Colonoscopy revealed one bleeding ulcer with raised margin (Fig. 2 A). Thus, biopsy specimens of the margin were obtained for the differential diagnosis of rectal cancer at 3 times. However, cancer cells were not found in every of the specimens, and hyperplasia of the crypt epithelium and fibromuscular obliteration were found in the specimens (Fig. 3). From these findings, the ulcer lesion was pathologically diagnosed as solitary ulcer syndrome of the rectum.

Because of his poor condition with severe anemia and diabetes mellitus, sigmoidostomy was performed on May 14 for resting the rectum. The macroscopic appearance of the ulcer changed gradually with time: the ulcer was covered with good granulation and the margin became flat 2 weeks after operation (Fig. 2 B). Anal bleeding had already stopped and the patient complained of no anal pain. Macroscopically, the ulcer became scar phase (Fig. 2 C) on June 9. The stoma was closed 4 weeks after operation on June 11, and the patients had uneventful postoperative course and discharged on July 10. The ulcer was completely cov-

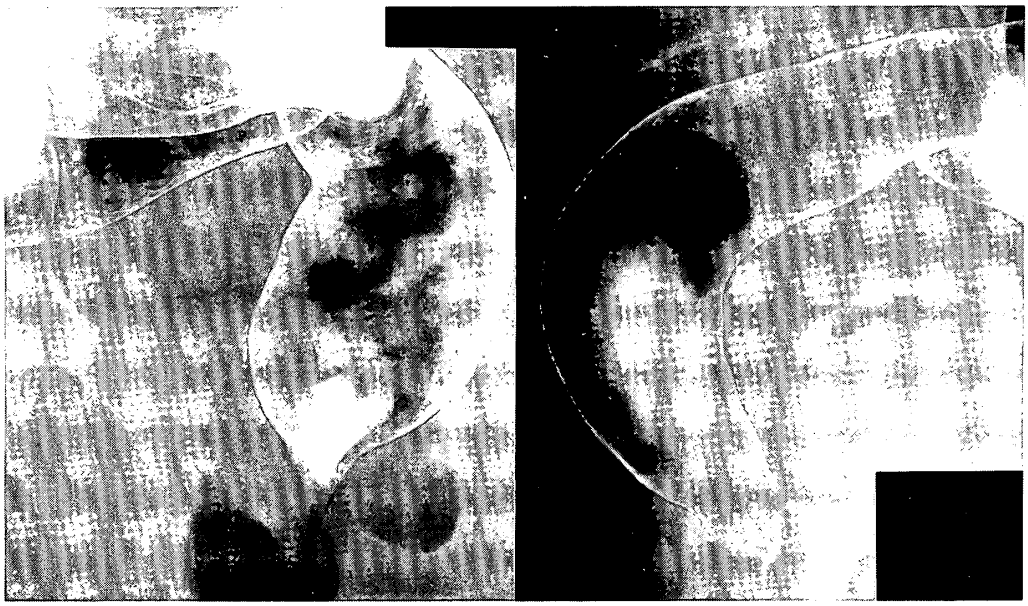

Figure 1. Barium enema of the distal bowel. A deep ulcer with a raised margin is clearly demonstrated in the anterior wall of the rectum.

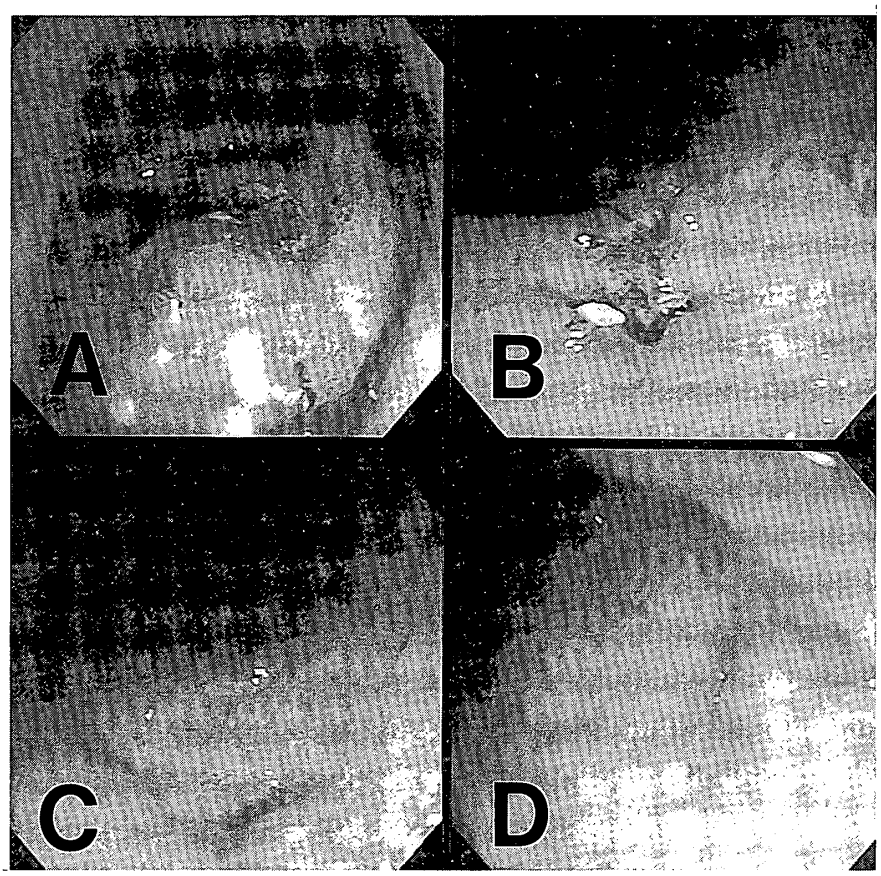

Figure 2. Colonoscopy of the rectum. A : deep ulcer with a raised margin on admission, B : the ulcer completely covered with good granulation and flattened margin before closure of sigmoidostomy, C : scar phase of the ulcer almost completely covered with epithelium just before discharge, D : the ulcer completely covered with epithelium 10 months after sigmoidostomy.

ered with epithelium 10 months after sigmoidostomy (Fig. 2 D). Thereafter, the patient took laxatives and avoided straining on defecation, and no recurrent sign was found 32 months after discharge.

Discussion

Solitary ulcer of the rectum is a rare disease ; only 19 case reports[5-12] have found in the Japanese literature for last 10 years. Since Madigan and Morson[1] have reviewed 68 cases of solitary ulcer of the rectum and described precisely its clinicopathologic characteristics in 1969, this ulcer has been widely accepted as one disease of the rectum[13]. The term "solitary ulcer of the rectum" is also used for this disease. However, Rutter and Riddel has proposed the term "solitary ulcer syndrome of the rectum" in 1975, because this disease is frequently complicated with various anorectal diseases and functions[4]. Thereafter, this term has been frequently used for this disease. In this case, external hemorrhoids were complicated.

In this disease, a considerable uncertainty has yet remained in pathogenesis. However, following explanation seems to be reasonable at the present time. A habit of difficulty on defecation followed

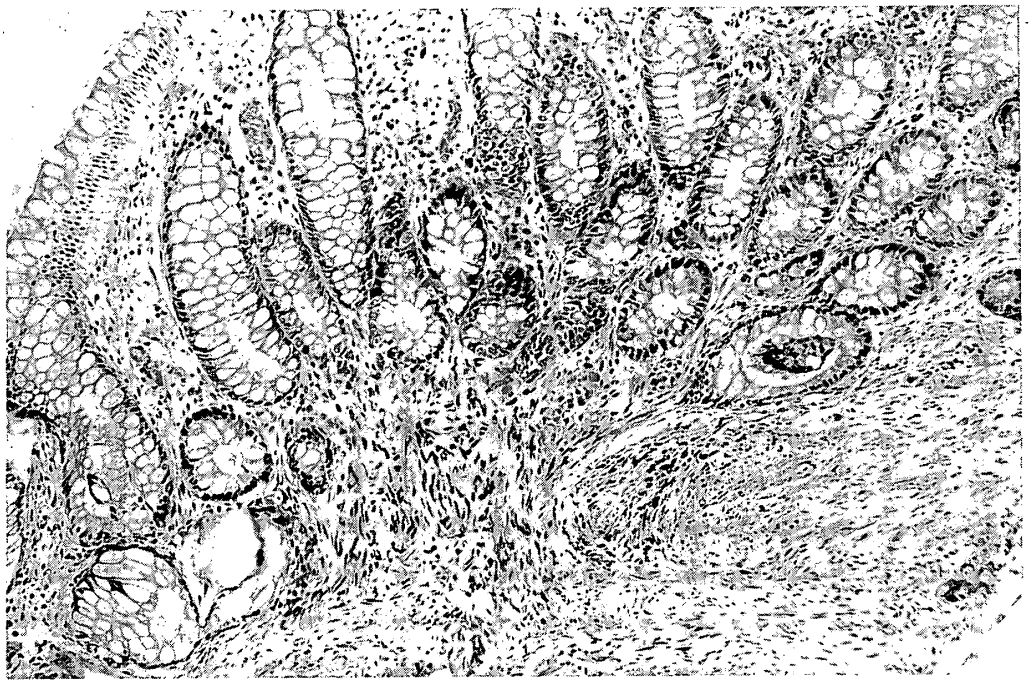

Figure 3. Microscopical photogram of the rectal biopsy specimen obtained from the ulcer margin. Hyperplasia of the crypt epithelium and fibromuscular obliteration of the laming propria are found. Original magnification × 100.

by straining is frequently associated with this ulcer[1-4, 8, 13-16]. The straining may lead to rectal mucosal prolapse, and the prolapse may induce trauma and ischemic change causing ulceration of the rectum[2, 8, 13-16]. The mucosal hyperplasia induced by the trauma and/or change may comprise nodular and/or polypoid lesions[2, 7, 13]. The failure of the puborectal muscle relaxation on straining is considered as a cause of this disease in some cases[3, 4, 8, 13]. In the present case, recurrence of the ulcer is not found after the correction of defecation without straining. Thus, solitary ulcer syndrome of the rectum seems to be caused by the just above mentioned phenomena.

Anal bleeding associated with mucous discharge and anal pain on defecation is the main symptoms[1-3, 8, 13-17]. This patient complained of anal pain and bleeding, and he had the aforementioned habit of strong straining on defecation. In general, common age of this disease has been reported be among the teens and fifties[1-3, 8, 13-17]. The present case is rare also in the point of age because of an aged patient. In many cases, the lesions has shown to be located within 15 cm from the anal margin and to involve commonly the anterior rectal wall[3, 8, 13-17]. The histopathological characteristics of this syndrome are : 1) proliferation of fibroblasts and smooth muscle in the stroma named fibromuscular obliteration by Madigan and Morson[1] ; 2) hyperplasia of the crypt epithelium ; and 3) ulcer or erosion. In some cases, villous configuration of the mucosa has been reported to be present[1-3, 8, 13-16]. In this case, the ulcer was present in the anterior wall of the rectum and accompanied with histopathological main 3 characteristics of this syndrome.

The first choice of therapies for patients with solitary rectal ulcer syndrome

is thought to be the conservative treatment, which includes a high roughage diets together with avoidance of straining on defecation and careful use of oral laxatives[1-3, 8, 13-17]. When the treatment has no or little effect on the symptoms and signs, surgical treatment is performed in general[1-3, 15, 16, 18]. Concerning surgical treatments, the methods have not yet been established. Local excision of the ulcer has been undergone in many cases[1-3, 8-13, 17], but Ford and co-workers have reported that the recurrence is found in almost half of the cases[3]. Abdominal and/or anteroposterior rectopexy may be recommended for this syndrome with rectal prolapse, as already reported some investigators[15, 16, 18]. In the present case, sigmoidostomy was performed for resting the rectum because of no effect of the conservative treatment and of aged patient with poor general condition. The recurrence is not found 32 months after discharge, although the patient has taken laxatives and avoided straining on defecation, as already suggested by Vaizey and co-workers[17]. Thus, colostomy may be one of the effective treatments, when the patient is in poor general condition.

References

1. Madigan MR, Morson BC : Solitary ulcer of the rectum. Gut 10 : 871–881, 1969
2. Rutter KRP, Riddell RH : The solitary ulcer syndrome of the rectum. Clin Gastroenterol 4 : 505–530, 1975
3. Ford MJ, Anderson HM, Gilmour S, et al. : Clinical spectrum of "solitary ulcer" of the rectum. Gastroenterol 84 : 1533–1540, 1983
4. Rutter KRP : Electromyographic changes in certain pelvic floor abnormalities. Proc Roy Soc Med 67 : 53–57, 1974
5. Wada H, Hirota H, Takahashi A, et al. : A case of solitary ulcer syndrome of the rectum. Chiryo 88 : 2007–2010, 1986 (in Japanese)
6. Kitatani H, Kawano M, Kajimoto T, et al. : Two cases of solitary ulcer syndrome of the rectum in children. Geka Shinnryo 29 : 1272–1273, 1987 (in Japanese)
7. Konishi H, Muto T, Adachi M, et al. : Solitary ulcer syndrome of the rectum with multiple polypoid lesions : report of a case. Stomach & Intestine 22 : 319–322, 1987 (in Japanese with English abstract)
8. Shibuya H, Huruya T, Nishida T, et al. : Studies on seven cases of solitary ulcer syndrome of the rectum. J Jpn Soc Clin Surg 49 : 337–343, 1988 (in Japanese with English abstract)
9. Mori M, Kashino H, Ishii H, et al. : A resected case of solitary ulcer syndrome of the rectum. Gastroenterol Surg 11 : 777–780, 1988 (in Japanese)
10. Shiota S, Nishi H, Yonekawa M, et al. : Three cases of solitary ulcer syndrome of the rectum. J Jpn Soc Clin Surg 25 : 1295–1300, 1990 (in Japanese with English abstract)
11. Sakaguchi M, Uehara K, Murakami Y, et al. : Three cases of solitary ulcer syndrome of the rectum. Ther Res 12 : 350–352, 1991 (in Japanese)
12. Kawaguchi Y, Nishikawa T, Maetani S, et al. : A case of solitary ulcer syndrome treated with transsacral proctomucosectomy. Jpn J Gastroenterol Surg 25 : 931–934, 1992 (in Japanese with English abstract)
13. Muto T : Solitary ulcer of the rectum. In Spectrum of Inflammatory Bowel Disease. Muto T (Ed.). Tokyo, Igaku-Shoin Co. Ltd., pp.139–145, 1986 (in Japanese)
14. Kuijpers HC, Schreve RH, Hoedemakers HC : Diagnosis of functional disorders of defecation causing the solitary rectal ulcer syndrome. Dis Col & Rect 29 : 126–129, 1986
15. Schweiger M, Williams JA : Solitary ulcer syndrome of the rectum. Its association

with occult rectal prolapse. Lancet 22 i : 170, 1977
16. Nicholls RJ, Simson JNL : Anteroposterior rectopexy in the treatment of solitary rectal ulcer syndrome without overt rectal prolapse. Br J Surg 73 : 222–224, 1986
17. Vaizey CJ, Roy AJ, Kamm MA : Prospective evaluation of the treatment of solitary rectal ulcer syndrome with biofeedback. Gut 41 : 817–820, 1997
18. Halligan S, Nicholls RJ, Bartram CI : Proctographic changes after rectopexy for solitary rectal ulcer syndrome and preoperative factors for a successful outcome. Br J Surg 82 : 314–317, 1995