

A Case of Ulcerative Colitis with Macroscopic Rectal Sparing

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= Abstract = A 35-year-old female patient, admitted with frequent mucoid bowel movements and lower abdominal pain, was suspected of having ulcerative colitis. She did not receive any treatment for ulcerative colitis prior to her admission. Despite the typical findings of ulcerative colitis in the transverse and descending colons on double-contrast barium enema and colonoscopy, the rectum was grossly normal. Such colonoscopic findings seemed to exclude the diagnosis of ulcerative colitis. However, rectal biopsy showed typical histologic changes of ulcerative colitis.

The purpose of this paper is to emphasize the importance of biopsy and histological assessment of the rectal mucosa in suspected cases of ulcerative colitis, even if the rectal mucosa is grossly normal on colono- or sigmoidoscopy.

Key Words: *Ulcerative colitis, Rectal sparing, Colonoscopy.*

INTRODUCTION

Chronic ulcerative colitis is a recurrent, inflammatory, and diffuse ulcerative disease of the colon and rectum which primarily involves the mucosa and submucosa (Kirsner, 1975; Hogan, 1980). While the cardinal features of chronic ulcerative colitis are diarrhea and rectal bleeding, the clinical manifestations are dependent upon the anatomical extent and intensity of the mucosal inflammation (Ritchie *et al.*, 1978). Ulcerative colitis was documented formally in 1875 by Wilks and Moxon. Thereafter, many reports of it have been made, and it became a not unusual disease in the Western world. But in Korea, some cases of ulcerative colitis have been reported, and it is considered a uncommon disease (Cho *et al.*, 1983; Park *et al.*, 1983; Kwon *et al.*, 1984; Kim *et al.*, 1985; Paik *et al.*, 1985; Han *et al.*, 1985; Ahn *et al.*, 1986; Yoon *et al.*, 1986; Ahn *et al.*, 1987; Han *et al.*, 1988;

Rhee *et al.*, 1990). Although the anatomical distribution of the inflammation on ulcerative colitis varies in many reports, the rectum is known to be involved in almost all cases. Thus, one should not diagnose ulcerative colitis in haste when the rectum is spared from the disease process. But in some cases of ulcerative colitis, where the rectum is apparently spared on endoscopic examination or radiologic study, rectal biopsies reveal the typical changes of ulcerative colitis (Jones, 1973; Burnham, 1980; Spiliadis, 1987).

This report describes a 35-year-old female patient who complained of frequent mucoid bowel movements and lower abdominal pain for six years. We had difficulty in diagnosing this case as ulcerative colitis because the rectum was grossly normal on not only double-contrast barium enema but on colonoscopy, despite the typical macroscopic changes of ulcerative colitis in the transverse and descending colons. So, we report this case with a review of the literature on rectal sparing ulcerative colitis.

CASE PRESENTATION

The patient under study was a 35-year-old woman who complained of having frequent mucoid bowel movements and lower abdominal pain for six years. The severity of the symptoms waxed and waned from the onset. She suffered from passing mucoid diarrhea up to 20 times daily while in aggravation. Two years prior to her admission, she was treated with anti amebic medication without alleviation of the symptoms. She did not receive any other specific treatment since then. About one month prior to admission, the frequency of mucoid diarrhea increased, which became four to five times a day at the time of admission. On physical examination, she was well-developed and moderately nourished. Blood pressure was 110/70 mmHg, respiration, 18/min, body temperature, 37 C (axilla). The conjunctiva was not anemic and the sclera not icteric. A small shallow ulceration was found on the tongue. No abnormality was found on the neck. Neither abnormality nor vasculitis was

noticed on fundoscopic examination. The chest and abdomen were negative except for an old operation scar on the lower abdomen. Tenderness, masses or mucoid stool were not noticed on digital rectal examination. The anal sphincter tone was normal. Laboratory studies were as follows: hemoglobin 12.2 gm%, leukocyte count 6,700/mm³, platelet count 316,000/mm³, on CBC. ESR was 15 mm/hr. Stools for ova, parasites, and heme were negative. Urinalysis was normal. VDRL and CRP were negative, and rheumatoid factor positive. Amebic titer was negative (below 1:8). HBsAg was negative. Serum protein and albumin were 7.0 gm%, and 3.7 gm%, respectively. Total bilirubin was 0.4 mg%. Transaminase, BUN/Cr, and serum electrolytes were normal. Chest P-A was normal.

Double-contrast barium enema revealed typical changes of ulcerative colitis, such as multiple pseudopolyps and loss of haustral markings along the transverse and descending colons (Fig. 1), but the rectum was grossly normal (Fig. 2). On colonoscopy, the rectum up to 20 cm above the anal verge was normal (Fig. 3), but from 20



Fig. 1. Air-contrast barium enema showing loss of haustral markings and multiple pseudopolyps from the transverse to descending colon.

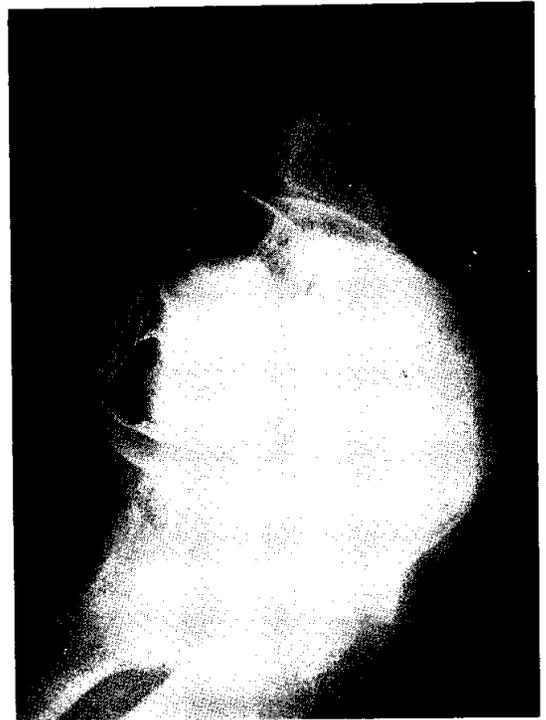


Fig. 2. Air-contrast barium enema showing normal rectum.

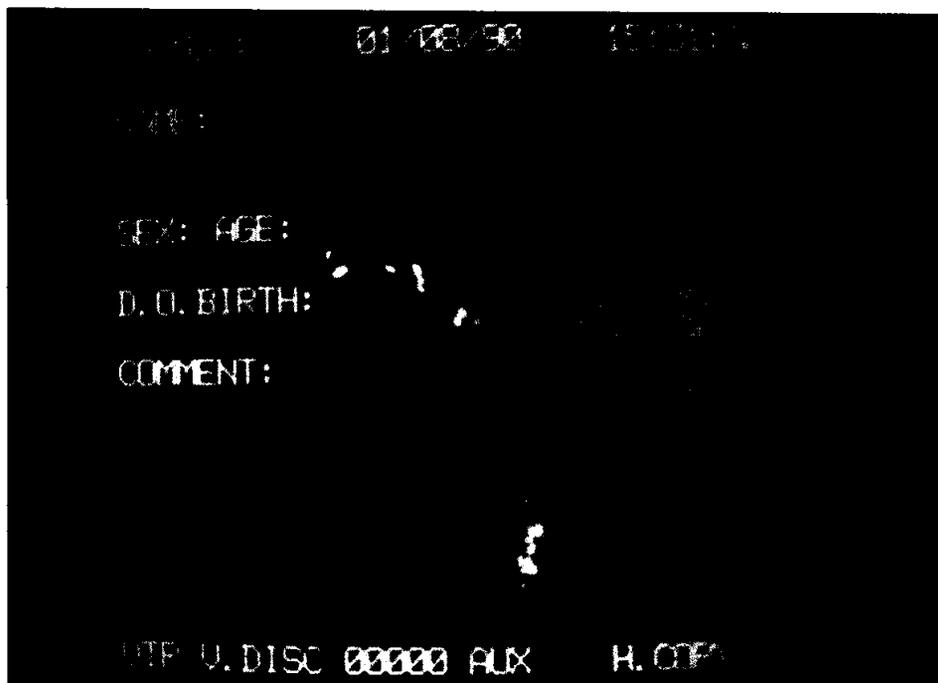


Fig. 3. Colonoscopic findings of the rectum showing grossly normal mucosa.

cm above the anal verge to the hepatic flexure, multiple pseudopolyposis, diffuse hyperemic mucosae, nonvisualization of submucosal vessels, and bleeding on touch were noticed (Fig. 4). Both colonoscopic biopsy specimens taken from the descending colon (Fig. 5) and from the normal looking rectum (Fig. 6) revealed consistent histologic changes of ulcerative colitis, such as depletion of the goblet cells, infiltration of the chronic inflammatory cells and neutrophils, decreased number of glands, distorted crypt architecture. But neither crypt abscess nor granuloma was found.

DISCUSSION

The extent of anatomical involvement in ulcerative colitis varies in many reports. The most common sites are the rectum or rectosigmoid colon (Ritchie *et al.*, 1978; Ahn *et al.*, 1987), the pancolitis (Kirsner *et al.*, 1975; Kwon *et al.*, 1984), the left-sided colon (Paik *et al.*, 1985; Rhee *et al.*, 1990), and the left colitis and proctitis (Ahn *et al.*, 1986). But there was no report of rectal sparing in ulcerative colitis in Korea. A number of uncontrolled studies have

already suggested that colonoscopy may be superior to barium enema in assessing the extent and severity of ulcerative colitis (Myren *et al.*, 1976; Elliott *et al.*, 1982). Colonoscopy with multiple biopsy is the procedure of choice in patients with proven or suspected inflammatory bowel disease (Holdstock *et al.*, 1984). Several reports have shown that routine barium enema underestimates the extent of colonic involvement and can even completely miss the total involvement of the colon (Myren *et al.*, 1976; Williams *et al.*, 1978).

The air-contrast barium enema is more reliable in revealing the extent of the disease. But still underestimates the extent in about 10-65% of the cases (Gabrielsson *et al.*, 1979). Of course, the barium enema has a role in the evaluation of the type of colonic contractility. Holdstock *et al.* (1984) reported that 18% of the patients were considered to have evidence of total colitis on barium enema, 38% on colonoscopy, and 61.8% on biopsy of their patients. Floren *et al.* (1987) also found as many as half of the patients with left-sided colitis on gross examination had total colitis histologically and suggested that the ana-



Fig. 4. Colonoscopic findings of the descending colon showing multiple pseudopolyposis and loss of submucosal vessels



Fig. 5. Microscopic findings of the descending colon.

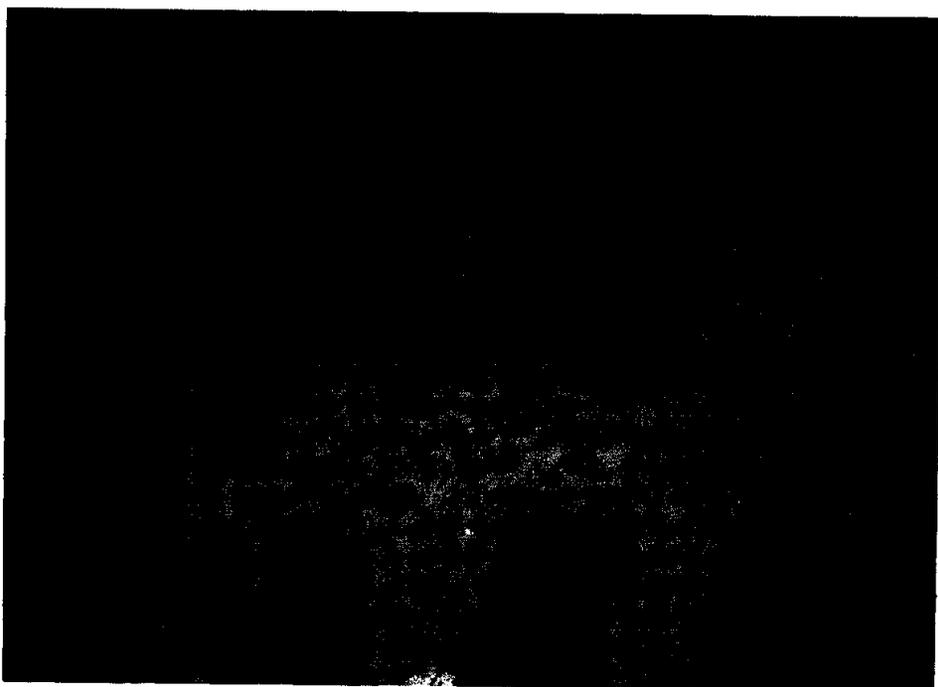


Fig. 6. Microscopic findings of the rectum revealed depletion of goblet cells, infiltration of the chronic inflammatory cells and neutrophils, and decreased number of glands. But neither crypt abscess nor granuloma was found.

tomical extent of the ulcerative colitis might often be underestimated endoscopically and that inflammatory activity could be present in normal looking mucosa on colonoscopy.

In typical ulcerative colitis, the colonic mucosa undergoes continuous, symmetrical inflammation. Endoscopically homogenous inflammatory changes, such as mucosal friability, ulceration, diffuse hyperemia, granularity, bleeding, erosions, pseudopolyps, and strictures are seen from the rectum to its proximal colonic side (Kirsner, 1975, Hogan *et al.*, 1980; Paik *et al.*, 1985).

The rectum is known to be involved in almost all cases of ulcerative colitis. However, Jones *et al.* (1973) reported that 5% of their patients with ulcerative colitis had a normal looking rectum on sigmoidoscopy, while others reported 3 to 4 percent. Burnham *et al.* (1980) suggested that the rectum may be spared in severe acute ulcerative colitis and concluded that sigmoidoscopic appearance is not a reliable indicator of extent in ulcerative colitis. But the number of patients

showing rectal sparing increases if relative sparing of the rectum (mild inflammation seen by endoscopy, with more severe proximal colonic lesions) is included in the definition. The rectum may be spared in a few patients with ulcerative colitis, especially during the healing process, by such means as locally administered corticosteroids (Williams *et al.*, 1981; Spiliadis *et al.*, 1987).

Although the rectum appears normal on endoscopy, histological examination always shows evidence of inflammatory changes, even if in remission (Frei *et al.*, 1982). Spiliadis *et al.* (1987) reported that rectal biopsies showed changes compatible with ulcerative colitis in all cases in analysis of a series of 12 patients with relative sparing of the rectum in ulcerative colitis. Thus, complete histologic sparing of the rectum was not observed.

Crohn's colitis is well-known for sparing the rectum in some patients (50%)(Jones *et al.*, 1973). One must differentiate Crohn's colitis from ulcerative colitis in patients in whom the rectum appears to be spared from the inflamma-

tion endoscopically. It is noteworthy that ulcerative colitis and Crohn's disease can be present simultaneously in the same patient (Otani, 1955; Steven *et al.*, 1980). On histopathologic examination, ulcerative colitis can be seen as nonspecific inflammation, ulceration, necrosis, granulation, crypt abscess, depletion of goblet cells, and submucosal edema (Paik *et al.*, 1985). In the most severe cases, the inflammation can involve the submucosa and even the serosa leading to perforation (Benard, 1988). Histological features that favor Crohn's disease include 1) focal as opposed to continuous inflammation in ulcerative colitis, 2) preservation of goblet cells, 3) lack of superficial ulceration, 4) submucosal or transmural inflammation, 5) large lymphoid follicles and lymphangiectasia, 6) eosinophils and macrophages in the lumen of the crypt abscess, 7) knife-like fissures, and 8) granulomas and microgranulomas (Steven *et al.*, 1988). In our patient, the rectum up to 20 cm from the anal verge was grossly normal, but its proximal site showed diffuse hyperemic mucosa, friability, and multiple pseudopolyps on colonoscopy. Microscopically, a decreased number of glands, infiltration of chronic inflammatory cells and neutrophils, and depletion of goblet cells were observed, but neither crypt abscess nor granulomas was found.

Although the rectum in ulcerative colitis may appear normal endoscopically, the rectal mucosa always shows evidence of disease on histologic examination, even though the inflammation may be focal (Spiliadis *et al.*, 1987). Nobuhide *et al.* (1989) suggested that topical administration of corticosteroids seems to have little therapeutic effect in patients with rectal sparing, contrary to the suggestion by Spiliadis *et al.* (1987) and that the relapse index was significantly higher in patients with rectal sparing.

Despite the low grade inflammation in the rectum preoperatively, colectomy with ileorectal anastomosis (rectal preservation) was occasionally unsuccessful in patients with rectal sparing ulcerative colitis, because some patients later required ileostomy due to postoperative proctitis, while others needed medical treatment for the inflammation of the remnant rectum (Spiliadis *et al.*, 1987).

Rectal sparing is uncommon in ulcerative colitis, even though the topical administration of

corticosteroids can mask the gross inflammatory change of the rectal mucosa. But in this case, the patient did not receive any treatment, such as administration of corticosteroids prior to admission. Colonoscopic biopsy specimens from the normal-appearing rectum also showed histological changes consistent with ulcerative colitis. We concluded that it is important to take biopsy specimens from the rectum and to assess the histologic involvement microscopically in suspected cases of ulcerative colitis, even if the rectal mucosa is grossly normal on colono- or sigmoidoscopy.

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= 국문초록 =

대장 내시경상 직장이 보존된 궤양성 대장염 1례

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남택서 · 김승민 · 이종관 · 정현제 · 송인성 · 최규완 · 김장봉

저자들은 6년 동안의 하복부 통증과 잦은 점액성 설사를 주소로 입원한 35세 여자에서 이중 대장 소장 조영술 뿐만 아니라 대장 내시경상으로도 횡행 결장과 하행 결장은 뚜렷적인 궤양성 대장염의 소견을 보임에도 불구하고 직장이 정상 소견을 보여 진단에 어려움을 겪었던 1례를 경험하였기에 문헌 고찰과 함께 보고하는 바이다. 대장 내시경상 정상 소견을 보인 직장 점막의 조직학적 소견은 출산 세포의 감소와 만성 염증성 세포의 침윤 등을 보여 궤양성 대장염에 해당하였다.

본 증례는 궤양성 대장염을 의심하는 경우에는 직장 점막이 대장 조영술 뿐만 아니라 대장 내시경상으로도 정상으로 보여도 반드시 조직 생검을 실시하여 병변의 조직학적 침범 여부를 관찰하는 것이 중요함을 강조하는 흥미있는 예로 생각된다.