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Clinicopathologic Comparison of Eroded Polypoid Hyperplasia and Solitary Rectal Ulcer Syndrome

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We experienced two unusual cases of tumor-like polypoid lesions involving the rectosigmoid colon. They could not be readily classified into any well known polypoid tumors of the rectosigmoid colon, but appeared to have some similarities to the previously documented "eroded polypoid hyperplasia (EPH)". A collective review of our seven cases of solitary rectal ulcer syndrome (SRUS), which proved to be due to paradoxically over-reactive muscle tone of the puborectalis, was performed, and clinicopathologic comparisons between EPH and SRUS were carried out. They shared histopathologic characteristics such as vascular congestion, crypt hyperplasia, and eroded surface, but they were different from each other in clinical symptoms, location of lesions and gross features. Furthermore, in one EPH case there was an altered mucin profile which was similar to that seen in SRUS and complete rectal prolapse. Conceivably, the pathological features of both EPH and SRUS were thought to have a possible connection with mucosal prolapse syndrome (MPS). Considering that MPS is a group of diseases encompassing SRUS and the related disorders of the colorectum and the anus, it is speculated that EPH of the rectosigmoid colon might be the proximal analogue of SRUS, a mucosal prolapse of the more distal colon.

Key Words : *Mucosal prolapse syndrome, Eroded polypoid hyperplasia, Solitary rectal ulcer syndrome, Rectosigmoid colon.*

INTRODUCTION

The polyposis syndrome of the rectosigmoid colon and anorectal junction have the characteristic anatomic location and intimate association with defecation resulting in mucosal prolapse (Boulay et al., 1983 ; Bogomoletz, 1992 ; Dean, 1993). MPS denotes a unifying, broad concept including SRUS(Schweiger, 1977 ; Boulay et al., 1983 ; Saul and Sollenberger, 1985 ; Chang YW et al., 1987) together with the related anorectal disorders such as inflammatory cloacogenic polyp (ICP) (Saul, 1987), colitis cystica profunda (CCP) (Bogomoletz, 1983) and even some nonproctologic conditions such as mucosal redundant polyp with diverticulosis (MRPD) (Franzin et al., 1985 ; Mathus-Vliegen and Tytgat, 1986 ; Kelly, 1991). We have experienced two cases of EPH, whose findings were comparable to those of SRUS and did not fit into any well-known polypoid disorders. This kind of polyposis in the rectosigmoid colon was once documented under

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the name of "eroded polypoid hyperplasia" (Burke and Sobin, 1990). This is to present the possible pathophysiogenesis as well as the morphological characteristics of this new entity with a comparative study of seven clinicopathologically proven cases of SRUS.

MATERIALS AND METHODS

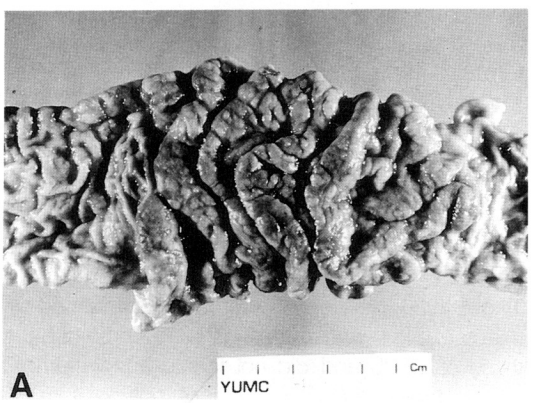
Presentation of two cases of eroded polypoid hyperplasia Case I

Clinical features

A 34-year-old male was experienced left lower abdominal pain and occasional mucoid diarrhea intermittently for 2 years. The patient's serum albumin decreased to 2.3g/dl. Repeated stool cultures, and serologic tests showed no evidence of infection. Sigmoidoscopic finding was multiple polypoid lesions in the 20cm proximal to the anal verge. There was a clinical suspicion of intestinal tuberculosis or malignant lymphoma, but the repeated sigmoidoscopic biopsies yielded "chronic nonspecific inflammation with erosion". Despite the lack of any firm evidence of malignancy on biopsy, the clinicians decided to resect the whole rectosigmoid colon to relieve the severe symptoms with signs and because of a high suspicion of malignancy.

Gross findings

A resected rectosigmoid colon demonstrated segmental polypoid lesions with sharp margins. The characteristics of the lesion were marked hypertrophic folds and gyric appearance with erosion of the mucosal surface. But there was no ulceration on the intervening remnant mucosa (Fig. 1A).



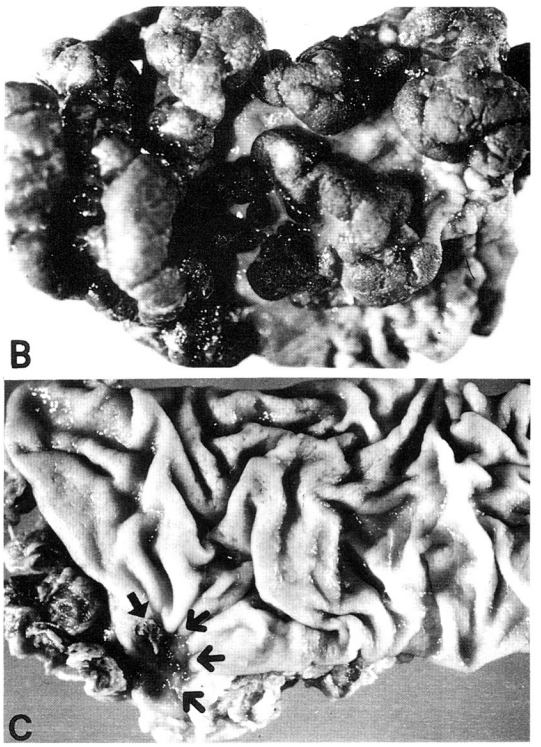


Fig. 1. Gross figures(A, B : EPH, C : SRUS) A : The involved segment shows hypertrophic mucosal folds with diffuse surface erosion, giving the superficial resemblance to cerebral gyri. B : A distinct lesion made of geographic, mulberry shaped folds with surface erosion. C : A sharply demarcated, punched-out ulcerative lesion(arrow) at the anterior wall of the rectum.

Pathological findings

Histopathologically the polypoid lesions demonstrated surface erosion, markedly tortuous and branched hyperplastic crypts with a deep proliferative zone and marked vascular congestion throughout almost the entire wall (Fig. 2).

Histochemistry

Mucin histochemistry revealed that sialomucin predominated over sulfomucin with Spicer's high iron diamine/alcian blue pH 2.5 (HID/AB) double stain (Spicer, 1965). Sulfomucin stained only the goblet cells lining the basal crypt.

Case II

Clinical features

A 38-year-old female complained of blood-tinged

Fig. 2. Polypoid lesion having a stalk which is composed of submucosa drawn by mucosal prolapse and hyperplastic crypts with deep proliferative zone. Note the congestion and vascular dilatation of the submucosa(H-E, ×1).

mucous diarrhea and complete rectal prolapse. Sigmoidoscopy demonstrated multiple polyps on the whole rectosigmoid colon. Several repeated biopsies revealed only "erosion with chronic nonspecific inflammation, granulation tissue formation, and regenerated crypts". The patient underwent mucosal stripping procedure (Delorme's operation) involving the rectosigmoid colon for reduction of the rectal prolapse.

Gross findings

The resected specimen showed mutually fused, geographically arranged, variable and thickened polyps with intervening mucosa of normal appearance. There was no ulceration. The surface of hypertrophic folds showed markedly shaggy, granular and erosive changes (Fig. 1B).

Histopathology and histochemistry

The histology and mucin profile were same as the former case.

Review of seven cases of solitary rectal ulcer syndrome

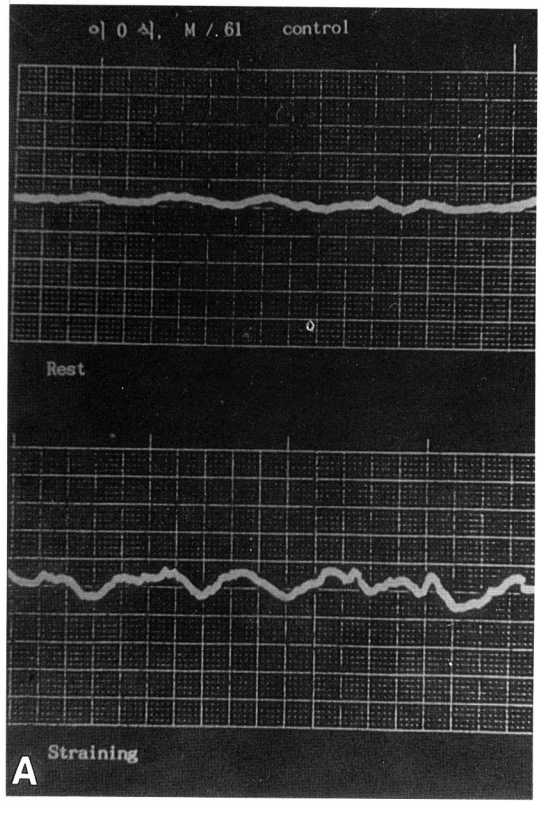
All seven cases were confirmed as SRUS by proctoscopic biopsy and manometry of the puborectal muscle at Severance hospital past during five years. In six of them, a biopsy was performed and the remaining one underwent segmental resection.

Clinical features

The patient's ages were all over 45 years, 4 of them were male. They complained of constipation, diarrhea with bloody stool and mild abdominal pain. All of them had the past histories including breast cancer, ovarian cancer, adenomatous polyp in the descending colon with tuberculosis and chronic liver disease. Only one patient had multiple ulcerative, erosive lesions and the remainder had a solitary ulcer on the anterior or antero-lateral wall of the rectum.

Manometry

Expressing sphincter activity as the mean of the puborectalis activities voiding in control subject was associated with inhibition of the sphincters to nearly resting level of activity (Fig. 3A). In SRUS patients voiding was accompanied by overreactivity of the sphincters that persisted throughout voiding (Fig. 3B).



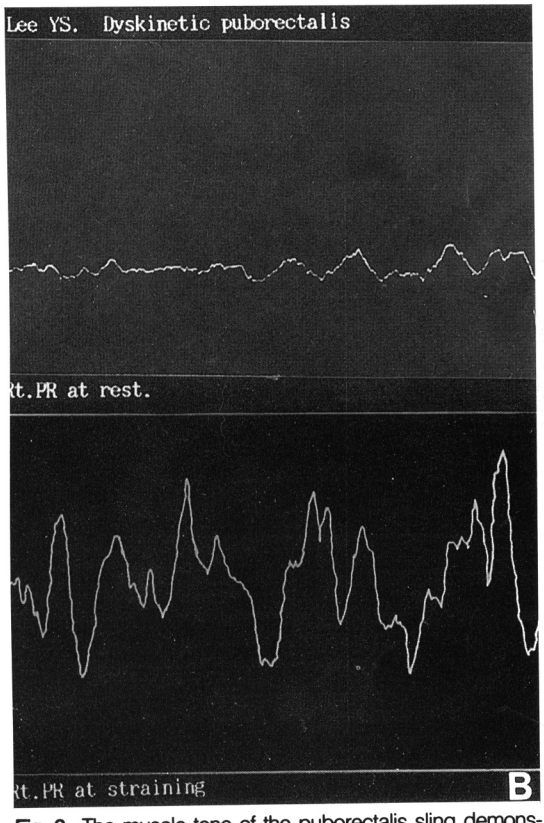


Fig. 3. The muscle tone of the puborectalis sling demonstrates paradoxically overreactivity during defecation by manometry(A : control, B : dyskinesia).

Gross findings

The resected rectum showed a sharply demarcated punched-out ulcerative lesion at the anterior wall of the rectum, measuring 1.7cm. The ulcer base was clear and its margin was flat(Fig. 1C).

Pathological findings

On sigmoidoscopic biopsy, common histological findings included mucosal ulceration with vascular congestion, hemorrhage and hypertrophy of muscularis mucosa. The other outstanding features were irregular hyperplastic branching of crypt with mucus hypersecretion and fibromuscular obliteration of lamina propria (Fig. 4A). Only one case demonstrated numerous scattered and grouped mucinophages (Fig. 4B).

Histochemistry

The mucin profile was also quite a similar to previous cases in that acid sialomucin predominated over acid sulfomucin.

Immunohistochemical findings

Immunoperoxidase stain for smooth muscle actin (monoclonal mouse anti-human α-smooth muscle actin primary antibody purchased from Dako Japan Co., Ltd.) was applied to all cases of SRUS, as well as EPH for the precise evaluation of ramifying branched fibromuscular obliteration of lamina propria from the thickened muscularis mucosa.

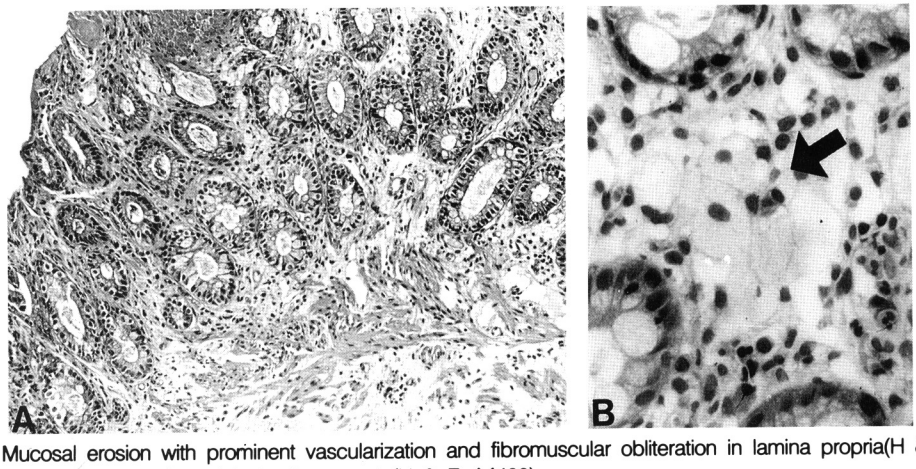


Fig. 4. A : Mucosal erosion with prominent vascularization and fibromuscular obliteration in lamina propria(H & E, ×100), B : Grouped mucinophages (arrow) in lamina propria(H & E, ×400).

Table 1. Comparison of eroded polypoid hyperplasia and solitary rectal ulcer syndrome in clinical settings

	EPH (2 cases)	SRUS (7 cases)
Age(yr)	34,38(mean : 36)	45-65(mean : 52)
Sex(M/F)	1/1	4/3
Site	rectosigmoid	anorectal junction
Number	multiple	single(6 cases)
Symptoms	severe	mild
	1 ; hypoalbuminemia	
	1 ; rectal prolapse	

EPH : Eroded polypoid hyperplasia.
SRUS : Solitary rectal ulcer syndrome.

RESULTS

The comparative results of both groups of EPH and SRUS were as follows : First of all in the clinical aspect (Table 1), EPH produced severe symptoms such as cramping pain, and hypoalbuminemia, while SRUS caused rather mild abdominal pain. The patients were younger in EPH (36) than in SRUS (52). In sigmoidoscopic findings, EPH cases showed multiple polypoid lesions throughout the rectosigmoid colon and SRUS showed a single ulcerative lesion in the anterior anorectal area except one case showing multiple lesions.

Both EPH cases aroused the clinical suspicion of malignancy, intestinal tuberculosis as well as Crohn's disease. Other common features of both EPH cases were as follows ; the age of onset in the 4th decade, severe symptoms such as hypoalbuminemia and complete rectal prolapse, clinically ominous suspicion of malignancy and the involvement of a specific site. Both shared the macro and microscopic features of hypertrophic, fused folds with segmental lesion, surface erosion, crypt hyperplasia and vascular congestion. Both had recovered after resection of the colons.

Table 2 demonstrated a comparison of EPH and SRUS in macro-and microscopic aspects. Each gross finding was quite distinct in that EPH showed multiple gyriform folds and almost all SRUS revealed an ulcer. Microscopically ulcerative evidence was noted at all cases of SRUS, while surface erosion instead of ulceration took place in two cases of EPH. Common sharing microscopic findings included crypt hyperplasia with vascular congestion and hypertrophy of muscularis mucosa. Fibromuscular obliteration of lamina propria was seen in minor cases of SRUS, whereas none of EPH.

Histochemically the mucin produced by the

Table 2. Comparison in gross and microscopic aspects

	EPH (2 cases)	SRUS (7 cases)
Gross finding		
gyriform folds	2	0
multiplicity	2	1
Microscopic finding		
mucosal ulceration	0	7
mucosal erosion	2	2
crypt hyperplasia	2	4
vascular congestion	2	4
MM* thickening	2	6
fibromuscular obliteration	0	2

EPH : Eroded polypoid hyperplasia
SRUS : Solitary rectal ulcer syndrome
* muscularis mucosa

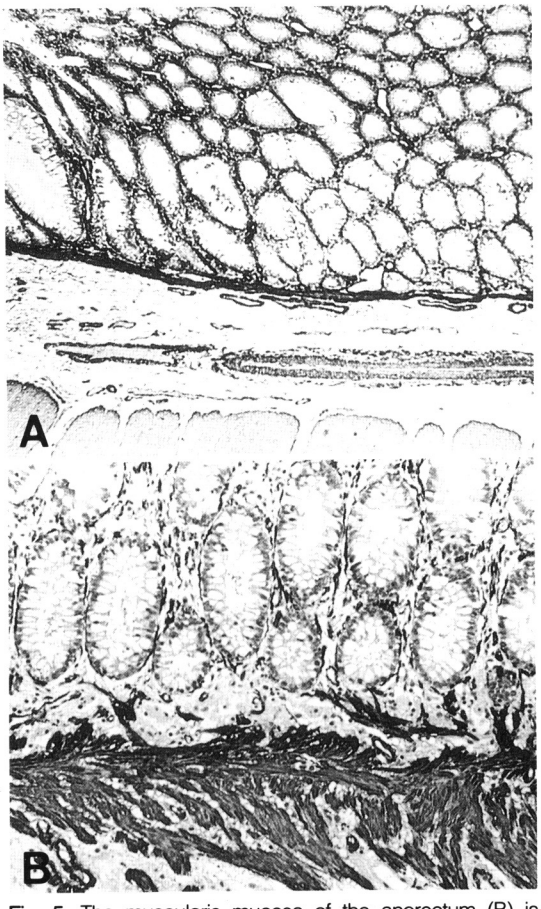


Fig. 5. The muscularis mucosa of the anorectum (B) is less continuous but thicker than that of the rectosigmoid colon (A) (LSAB for smooth muscle actin, ×40).

glands in both conditions of SRUS and EPH showed to be abnormal, with sialomucin predominance and scanty sulfomucin only in basal crypts.

Immunohistochemical method using actin made reaffirmed that there was a big difference between the nature of muscularis mucosa of anorectal junction and rectosigmoid colon. The muscularis mucosa in EPH (Fig. 5A) was less discontinuous and splintered than that of SRUS (Fig. 5B).

DISCUSSION

We experienced 2 cases of unusual and unclassified polyposis in the whole rectosigmoid colon, very similar to EPH previously documented by Burke and Sobin (Burke and Sobin, 1990). They have said that EPH newly named would be peculiarly characterized by severe symptoms such as hypoalbuminemia, severe abdominal pain and the involvement of the specific site, only at the rectosigmoid colon. EPH also has the histopathological finding very similar to those of MPS except that it is neither associated with ulceration nor fibromuscular obliteration.

Our cases of SRUS demonstrated a paradoxically overreactive muscle tone by manometry on the puborectal muscle sling. The paradoxical contracture tone by barodamages makes the mucosa prolapse into more distal side in accordance with several minor factors such as mechanical trauma by hard stool. Because of the antero-lateral wall of the rectal mucosa being the nearest to the abdominal wall, in SRUS it is quite labile to a high transmural pressure gradient during defecation (Womack et al., 1987). This results in local ischemic changes in the region which finally result in ulcer. This hypothesis has been widely accepted recently and applied to several diseases including SRUS, ICP and MRPD. Some authors, therefore, insist on unifying these diseases as MPS (Boulay et al., 1983).

MPS has been accepted as a unifying clinicopathologic concept for SRUS with related anorectal condition ; showing SRUS-like changes such as ICP and MRPD in even more proximal sites (Boulay et al.,1983 ; Bogomoletz, 1992). These divergent entities, however, share the common histologic features, and then they might be classified into a single category. From a theoretical viewpoint, distal area of the sigmoid colon is downward oriented and closely connected with defecation, and in turn always susceptible to high intravisceral pressure with an additive effect of intrinsic muscle as well as sphincteric muscle contraction (Rutter, 1974 ; Womack et al., 1987).

From a pathological viewpoint, the common features of MPS are so remarkable as to be a crypt hyperplasia and ulceration as well as vascular congestion with fibromuscular obliteration (Schweiger, 1977 ; White et al., 1980 ; Boulay et al., 1983 ; Saul and Sollenberger, 1985 ; Womack et al., 1987 ; Sun et al., 1989 ; Williams et al., 1991 ; Bogomoletz, 1992 ; Dean, 1993 ; Lonsdale, 1993).

Clinicopathologic features of our cases of EPH were the same as those of the above-mentioned cases (Burke and Sobin, 1990). As in Table 1, the mean age of onset of EPH patients was lower than that of SRUS patients. This is somewhat different

from other reports in which the peak age in SRUS was the 3rd or 4th decade. EPH had a tendency for occurring in the rectosigmoid colon as a predilection site and for multiplicity. On the contrary SRUS was almost always a single lesion in the anorectal junction. Both cases of EPH showed severe symptoms. Besides both were of an ominous nature. These led to the tentative diagnosis of tumor or inflammatory bowel disease such as localized Crohn's disease or intestinal tuberculosis. Despite the clinical course and findings suggestive of tumor or inflammatory bowel disease limited to the rectosigmoid colon, the histological findings of the biopsied specimen were not typical of any well-known disease, but very similar to those of MPS. We found, however, the prominent diffuse surface erosion even in nearly normal-fold thickened mucosa. We also found vascular congestion simulating angiodysplasia with crypt hyperplasia like a hyperplastic polyp and mucous hypersecretion.

Whereas almost all cases of SRUS showed mild symptoms and made rather an accurate diagnosis possible due to small sharply demarcated ulcerative lesion on anorectal junction. In pathological view of SRUS, some findings including ulceration instead of erosion and occasional fibromuscular obliteration of lamina propria were absolutely different from cases of EPH. These different points between SRUS and EPH in macro and microscopical aspects were very interesting and intriguing.

The fact that only erosion not ulceration has occurred in cases of EPH could be considered as an inevitable result from the well-known functional vascular pathophysiological process (Boley et al., 1977). According to them, vascular dilation might be of congestion rather than a malformation, the major contributing factor being simply a repeated muscular over-contraction. The capillary system of the tip of the colonic mucosa would be the most susceptible to ischemia. The repeated erosion without ulceration, despite massive crypt hyperplasia, is still to be clarified. This fact which is not associated with any ulcer may be closely related with the peculiar anatomical characteristics of the rectosigmoid colon. It is well known that the rectosigmoid colon is more loosely attached with mesentery and has wider lumen than the anorectal junction (Burke and Sobin, 1990). These differential points may explain why only the erosive change occurs in EPH and may predispose less to ulcer than anorectal junction.

Another remarkable differential point of both groups in histological findings is a lack of insinuation of muscle fiber splintered from muscularis mucosa into the overlying lamina propria, so called fibromuscular obliteration in cases of EPH. Burke and Sobin also explained this event by the native characteristics of muscularis mucosa (Burke and Sobin, 1990). This study confirms that the muscularis mucosa of normal anorectum is less continuous but thicker than that of the rectosigmoid colon.

In my opinion, several hypotheses for underlying mechanisms could be put forward in the pathogenesis of this new entity, EPH.

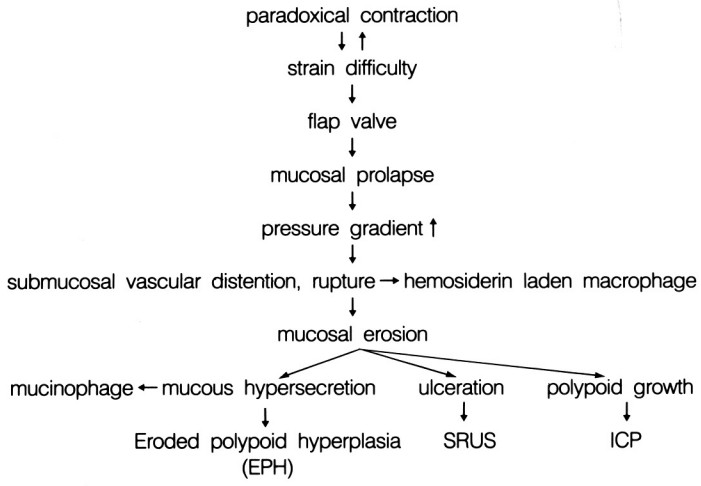
First of these is a concept that surface erosion follows crypt hyperplasia. The crypt hyperplasia occurs as if a hyperplastic or hamartomatous polyp does. The following event is a vascular insufficiency which could be resulted from the stretched submucosal and mucosal vasculature. The next sequence is surface erosion as a result of vascular insufficiency. Finally inflammatory process and granulation tissue may be formed as a reaction of surface erosion. But this hypothesis is not satisfactory to explain the erosive area even in mucosa of normal thickness, and vascular congestion of the submucosa. Additionally the hyperplastic polyp would be scarcely, if ever, associated with surface erosion.

The second hypothesis is the concept that crypt hyperplasia follows surface erosion. The compensatory crypt hyperplasia with inflammation and granulation tissue follows the erosion due to barodamage or mechanical trauma. The remarkable changes of submucosa, however, can not be explained with satisfaction even in this hypothesis.

The third hypothesis is crypt hyperplasia and erosion follow the mucosal prolapse. The mucosal prolapse from occult to overt, by the above mentioned mechanism, may be the predisposing factor to the following sequence. Only this mechanism can give an explanation for the changes of submucosal vascular congestion and dilation.

In an overview, there is a vicious cycle between the paradoxical contraction and straining difficulty which results in a flap valve and then an eventual prolapse with aggravation by additive effects of the local stercoral damage. The mucosal prolapse increases transrectal pressure gradient which in turn mucosal erosion directly as well as distention and rupture of submucosal vasculature. At that time, hemosiderin laiden macrophage or hemorrhage may be seen in the submucosa. The resultant

Table 3. Sequential steps of the morphogenesis of eroded polypoid hyperplasia



SRUS : Solitary rectal ulcer syndrome.
ICP : Inflammatory cloacogenic polyp.

mucosal erosion continues in a repeated manner, spilling over mucin which is taken up eventually by mucinophages to appear as in xanthoma (Table 3). We can not find microthrombus in any vasculature, previously suggested to be a major contributing factor of MPS (Lonsdale, 1993). The majority of studies on the patterns of mucin secretion in benign colonic polyp until the mid-1980's concluded that there were few clues for differentiating the variable entities of benign polyps (Listinsky and Riddell, 1981 ; Franzin et al., 1983 ; Franzin et al., 1984 ; Jass et al., 1984). All of the reviewed cases showed sialomucion predominance over sulfomucin detected only in basal crypt, as in a so called "transitional mucosa" adjacent to large bowel tumor. HID/AB stain has revealed a preponderence of sulfomucin over sialomucin in hyperplastic or juvenile retention polyp (Listinsky and Riddell, 1981 ; Franzin et al., 1983 ; Franzin et al., 1984 ; Jass et al., 1984). Essentially we found the differential staining pattern between EPH and other polyps.

We would conclude that EPH was a morphologic variant,site-specific, of MPS quite similar to SRUS in morphological and functional aspects.

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