# Seromuscular Tear of the Sigmoid Colon in the Absence of Trauma: A Case Report

Tsutomu Nomura, Masao Miyashita, Itsuro Fujita, Hideyuki Suzuki, Kiyonori Furukawa and Takashi Tajiri

Surgery for Organ Function and Biological Regulation, Graduate School of Medicine, Nippon Medical School

## Abstract

A 16-year-old adolescent boy was admitted to our hospital with severe lower abdominal pain and was found to have peritonitis, probably caused by acute appendicitis. At laparotomy, we found a paper-thin, dilated sigmoid colon; the seromuscular layer on the antimesenteric side was torn, and the untorn mucosa showed a pinpoint perforation. The seromuscular defect had spread circumferentially to involve the entire circumference of the colon wall. We performed sigmoidectomy, and the patient recovered uneventfully. This case showed many similarities, both in terms of the macroscopic and pathological findings, to seromuscular tear, an entity specifically associated with seatbelt use. This case is noteworthy because seromuscular tear-like lesions of the colon without a history of trauma has not previously been reported. (J Nippon Med Sch 2008; 75: 289–292)

Key words: seromuscular tear, intraluminal dissociation of intestine, spontaneous perforation of colon

#### Introduction

A morbid state characterized by dissociation of the mucous membrane and the muscle layer of the alimentary tract is extremely rare. We report a case of such intraluminal dissociation and pinpoint perforation of the sigmoid colon without a history of trauma. Both macroscopically and pathologically, this case showed many similarities to seromuscular tear (SMT), an entity specifically related to seatbelt use<sup>12</sup>.

We believe this case is noteworthy because a colon tear resembling SMT, in terms of either the gross findings or the pathological findings, in the absence of trauma has not previously been reported.

## **Case Report**

A 16-year-old adolescent boy was admitted to our hospital with severe lower abdominal pain. Laboratory examination revealed elevation of the white blood cell count (15,500 /mm<sup>3</sup>) and the serum C-reactive protein (7.3 mg/dL); however, all other results, including hematology, test general biochemistry, and urinalysis, were within normal limits. Physical examination revealed rebound tenderness in the lower abdomen, and a diagnosis of peritonitis, probably due to acute appendicitis, was made. At laparotomy, there was evidence of peritonitis, but the appendix appeared normal.

Correspondence to Tsutomu Nomura, Department of Surgery, Nippon Medical School, 1-1-5 Sendagi, Bunkyo-ku, Tokyo 113-8603, Japan E-mail: nomura-t@nms.ac.jp

Journal Website (http://www.nms.ac.jp/jnms/)

#### T. Nomura, et al



Fig. 1 Operative findings. The intestine without the seromuscular layer ( \* ) was found to have herniated through the SMT (arrow).



Fig. 2 Macroscopic findings of the specimen.a: Mucosa-submucosa bridge spanning the tear (\*).b: The SMT was approximately 10 cm long on the antimesenteric side (arrows).

Instead, we found a paper-thin, dilated sigmoid colon with a tear of the seromuscular layer on the antimesenteric side, and the untorn mucosa showed a pinpoint perforation. The seromuscular defect had spread circumferentially to involve the entire circumference of the colon wall. The defect was approximately 10 cm long on the antimesenteric side, and the intestine without the seromuscular layer was found to have herniated through the tear (Fig. 1). There was a large amount of stool in the sigmoid colon; however, no fecal contamination of the abdominal cavity had occurred because the perforation of the mucosa was extremely small and the stool was extremely hard. We performed sigmoidectomy, and the patient recovered

uneventfully.

The macroscopic findings of the specimen are shown in **Figure 2**. Pathological examination revealed dissection between the submucosal layer and the muscular layer (**Fig. 3a**). Edema of the submucosal layer was observed in association with ectasia and congestion of the submucosal veins (**Fig. 3b**, c).

## Discussion

To discuss the pathogenesis of the morbid condition in this patient, we searched the literature for similar cases. However, there have been few reported cases of dissociation between the muscle



Fig. 3 Pathological findings of the specimen.a: Dissection between the submucosal layer and the muscular layer was observed.

- **b**: The wall of paper-thin intestine. Mucosasubmucosa layer without a seromuscular layer was observed.
- c: Edema of the submucosal layer ( \* ), ectasia, and congestion of the submucosal veins (arrow) was observed.

layer and the mucous membrane inside the wall of the alimentary tract. We found a few reports of cases of dissection occurring due to barium enema<sup>34</sup> and of intraluminal hematoma due to trauma<sup>5</sup> Another review has suggested that dissociation is a complication of colonic diverticulum<sup>6</sup>. However, our case had no features in common with previously reported cases. In particular, the patient had not sustained an injury and had not undergone barium enema.

Ehlers-Danlos syndrome type IV, a inheritable disease of type III collagen metabolism, may cause spontaneous colonic perforation. However, the clinical findings and family history of the present case did not meet the criteria of this disease<sup>78</sup>.

Pathological findings are most important for determining the cause of this rare disease. A large defect of the serosa and muscle, probably due to the tear of the seromuscular layer, was observed, and the leading edge of the tear stripped the inner muscularis from the submucosa. On the other hand, a "submucosal-mucosal bridge" was found spanning the tear. These pathological characteristics are strikingly similar to those of the circumferential type of SMT, which was reported by Garret and Braunstein to be a component of seatbelt syndrome in 19629; the hallmark intestinal injury of seatbelt syndrome is caused by a tear that separates the inner muscularis from the submucosa. Slavin et al. have reviewed the features of SMT<sup>10</sup> on the basis of the clinical and pathologic findings in 29 patients who underwent emergent operation after motor vehicle accidents. SMT occurred in 90% of their patients, with 73% of all SMTs developing in the colon and one-third occurring in the sigmoid colon. They reported the following characteristic features of SMT: 1) a wedge that strips the submucosa from the inner circular muscle; 2) a bending retraction of the torn muscularis towards the uninvolved bowel wall; 3) mucosal-submucosal fold effacement, causing the mucosa-submucosa bridge spanning the tear to become paper thin; and 4) vulnerability of this bridge to ischemia and that 35% of the tears studied culminate in incipient or frank perforations or gangrene or both. Slavin et al. have concluded that SMT is caused by the simultaneous activation of different traumatic mechanisms on the bowel and its mesentery in persons wearing a seatbelt and involved in motor vehicle accidents. However, our patient did not have a history of trauma; accordingly we cannot use the same explanation as for SMT to explain his morbid condition.

Because a 10-cm-long complete dissociation was found at operation, some kind of physical force might have acted on that area. This force was most likely to be the internal pressure of the intestine. In this case, the dissociated part was markedly dilated, and a large volume of stool was observed in the sigmoid colon which could have markedly increased the internal pressure. However, it is difficult to imagine such dissociation as in this case to have been caused by only an increase in internal pressure. We hypothesize that that seromuscular defect of the sigmoid colon, for example, a diverticulum or small SMT caused by unknown factors, might have been present in our patient. Increased internal pressure of the sigmoid colon and weakness of the colonic wall, peristalsis, and other factors may have played a role in the development of this morbid state of the sigmoid colon.

#### References

 Appleby JP, Nagy AG: Abdominal injuries associated with the use of seatbelts. Am J Surg 1989; 157: 457– 458.

- Williams JS, Kirkpatrick JR: The nature of seat belt injuries. J Trauma 1971; 11: 207–218.
- Cho KC, Simmons MZ, Baker SR, et al.: Spontaneous dissection of air into the transverse mesocolon during double-contrast barium enema. Gastrointest Radiol 1990; 15: 76–77.
- Greves JH 3rd, Bohlman TW, Frische LH, et al.: Intramural barium in ischemic colitis: a new radiographic finding. Am J Dig Dis 1976; 21: 257–262.
- Calabuig R, Ortiz C, Sueiras A, et al.: Intramural hematoma of the cecum: report of two cases. Dis Colon Rectum 2002; 45: 564–566.
- Stein GN: Radiology of colonic diverticular disease. Postgrad Med 1976; 60: 95–102.
- Beighton P, De Paepe A, Steinmann B, et al.: Ehlers-Danlos syndromes: revised nosology, Villefranche, 1997. Ehlers-Danlos National Foundation (USA) and Ehlers-Danlos Support Group (UK). Am J Med Genet 1998; 77: 31–37.
- Watanabe A, Kosho T, Wada T, et al.: Genetic aspects of the vascular type of Ehlers-Danlos syndrome (vEDS, EDSIV) in Japan. Circ J 2007; 71: 261–265.
- Garrett JN, Braunstein PW: The seatbelt syndrome. J Trauma 1962; 2: 220–238.
- Slavin RE, Borzotta AP: The Seromuscular Tear and Other Intestinal Lesions in the Seatbelt Syndrome: A Clinical and Pathologic Study of 29 Cases. Gut 2002; 23: 214–222.

(Received, April 25, 2008) (Accepted, May 23, 2008)