A case of colon lymphangioma treated with laparoscopy-assisted ileocecal resection

Kazuhito Nabeshima*, Takao Machimura**, Mitsuru Wasada***, Kyoji Ogoshi*, Hiroyasu Makuuchi*,

*Department of Surgery, Tokai University School of Medicine, **Department of Surgery, ***Department of Internal Medicine, Ikegami General Hospital

(Received January 10, 2008; Accepted February 28, 2008)

We encountered a patient with colon lymphangioma and performed laparoscopy-assisted ileocecal resection. The patient was a 68-year-old male who visited our hospital for weight loss in May 1999. Since fecal occult blood was positive on close examination, colonoscopy was performed. A light-permeable, transparent, and pedunculated polyp was found in the ascending colon and diagnosed as submucosal tumor. The patient was admitted for close examination and treatment. Abdominal CT detected a tumor in the ascending colon accompanied by disturbed surrounding adipose tissue, suggesting extramural invasion of the tumor. Based on the diagnosis of extramural submucosal tumor of the ascending colon, ileocecal resection was performed with laparoscopic assistance. The laparoscopic findings were as follows: 1) Lymph vascular dilatation filled with lymph expanding on the serosal surface, 2) light-permeability/translucency, 3) laparoscopic cushion sign, and 4) lymph vascular dilatation in the surrounding adipose tissue. Laparoscopy-assisted surgery was useful for the diagnosis and treatment of colon lymphangioma, and characteristic laparoscopic findings were noted.

Laparoscopic surgery is a useful for a diagnosis and the treatment of large lymphangioma of colon.

Key words: Colon lymphangioma, laparoscopic surgery, ileocecal resection

INTRODUCTION

Lymphangioma is a benign tumor generally occurring in children. The cause of lymphangioma is congenital malformation of the lymphatic system. Lymphangioma generally develops in the head and neck region and axilla, but rarely develops in the abdominal cavity. Colon lymphangioma is a relatively rare non-epithelial tumor. We encountered a patient with colon lymphangioma and performed laparoscopy-assisted ileocecal resection. We report the case with a literature review because characteristic laparoscopic findings were noted.

CASE REPORT

A 68-year-old Japanese man was admitted to Ikegami General Hospital in May 1999. He complained loss of weight of 15kg in 3 months from May 1999. Since fecal occult blood was positive, colonoscopy was performed, and a pedunculated polyp was noted in the ascending colon. The abdomen was soft and flat, and no superficial lymph node was palpable.

No abnormalities were noted in general blood test, blood chemistry, or urinalysis. Blood tumor marker levels were within the normal ranges.

Colonoscopy revealed was a pedunculated polyp with a smooth surface was observed directly above the Bauhin valve of the ascending colon. The tumor was ash-colored, compared to the surrounding, with no marked mucosal change, suggesting submucosal tumor. The submucosal tumor was light-permeable and translucent (Fig. 1).

Contrast enema revealed a filling defect with a clear margin was noted in the ileocecal region (Fig. 2).Abdominal CT revealed a tumor with low density content was found in the ascending colon. The condition of the surrounding adipose tissue suggested extramural invasion of the tumor (Fig. 3).Based on the diagnosis of submucosal tumor of the ascending colon, laparoscopy-assisted ileocecal resection was performed. Surgical findings were swelled translucent lymph vessels noted on the serosal surface of the ascending colon by laparoscopy. When the lesion was compressed with forceps, an endoscopic cushion sign-like soft touch was felt. Similar lymph vascular dilatation was also found in the outer adipose tissue of the ascending colon (Fig. 4 a,b,c). Excised specimen was dilated and protruded lymph vessels were noted on the serosal surface of the ascending colon. A tumor covered with normal mucosa was observed in the ascending colon directly above the Bauhin valve (Fig. 5).

Histopathological findings was the tumor mainly located in the submucosal layer, and dilated lymph vessels expanded from the mucosal to subserosal layers, in which lymph was filled. Based on these findings, colon lymphangioma was diagnosed (Fig. 6).

The post operative course of this patient was uneventful.

DISCUSSION

Colon lymphangioma is a non-epithelial tumor that develops in the colon, and the incidence is very low.

Kazuhito Nabeshima, Department of Surgery, Tokai University School of Medicine, 143 Shimokasuya, Isehara, Kanagawa, Japan, 259-1193,
Tel: +81-463-93-1121, Fax: +81-463-95-6491, E-mail: nabesima@is.icc.u-tokai.ac.jp

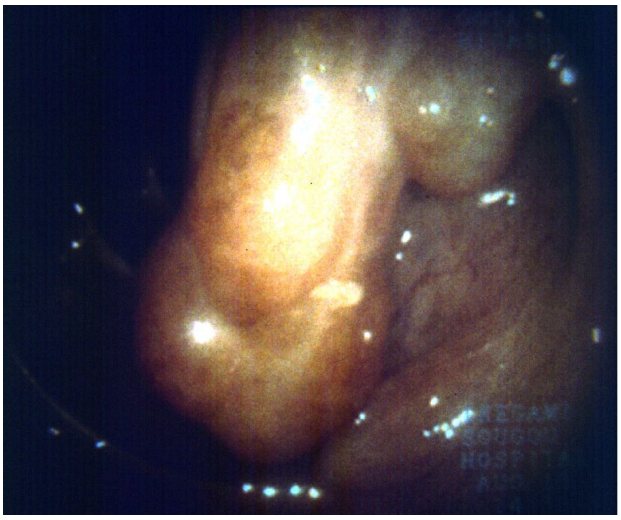

Fig. 1. Colonoscopic findings: A smooth-surfaced pedunculated polyp was noted directly above the Bauhin valve of the ascending colon.

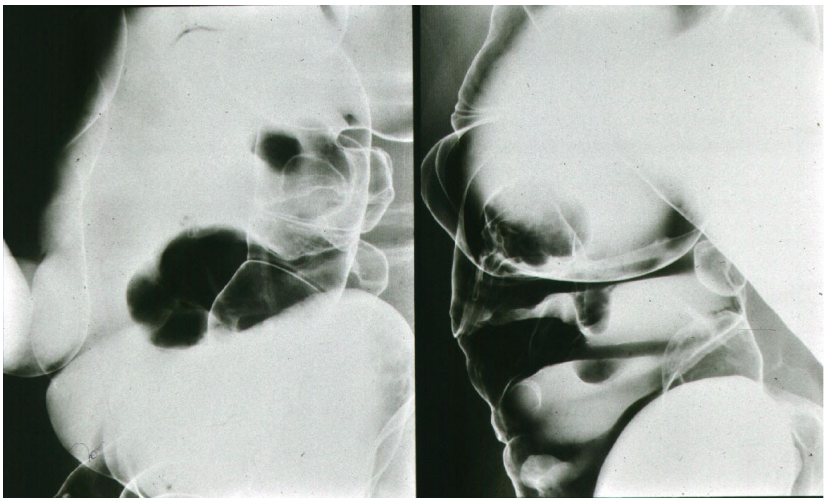

Fig. 2. Contrast enema findings: A filling defect with a clear margin was noted in the ileocecal region.

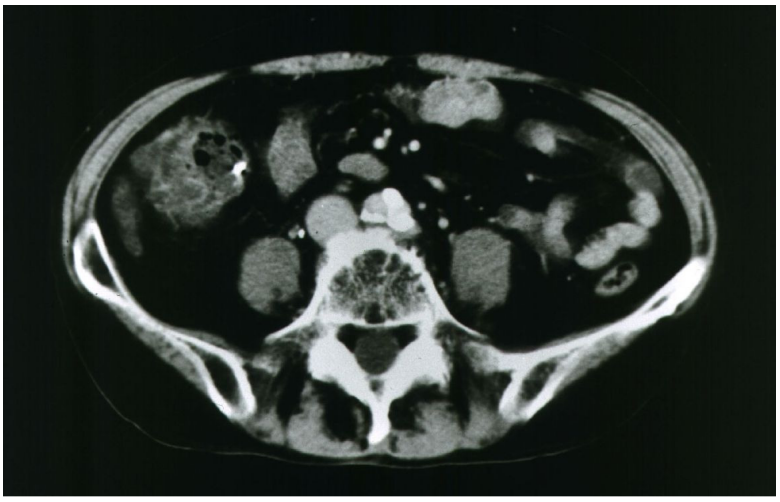

Fig. 3. Abdominal contrast CT findings: A tumor with a low- to water-density content was present in the ascending colon.

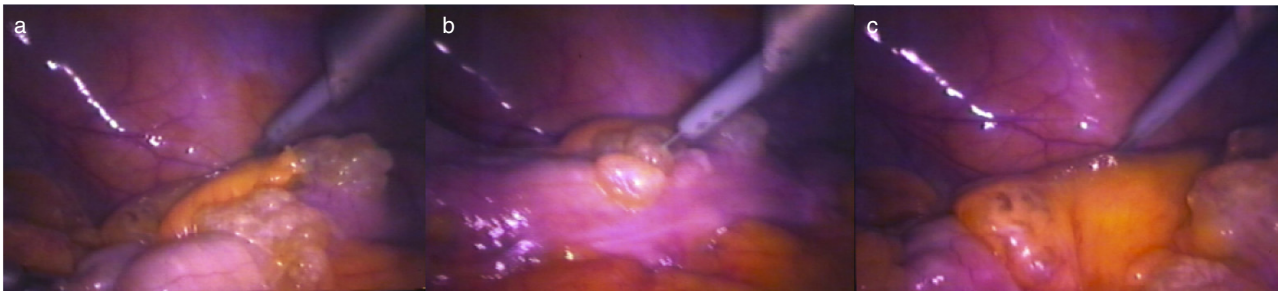

Fig. 4. a,b,c: Laparoscopic findings: a) A swelled transparent lymph vessels wee noted on the serosal surface of the ascending colon. b) A soft touch was felt when the tumor was compressed with forceps. c) Similar lymph vascular dilatation was noted in the outer adipose tissue of the ascending colon.

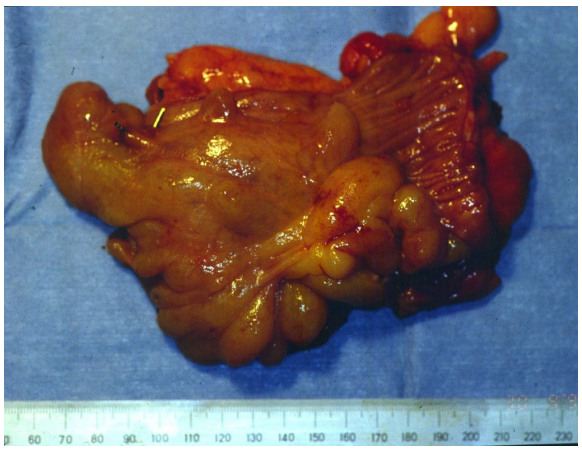

Fig. 5. Findings in excised specimen: A tumor covered with normal mucosa was present in the ascending colon directly above the Bauhin valve.

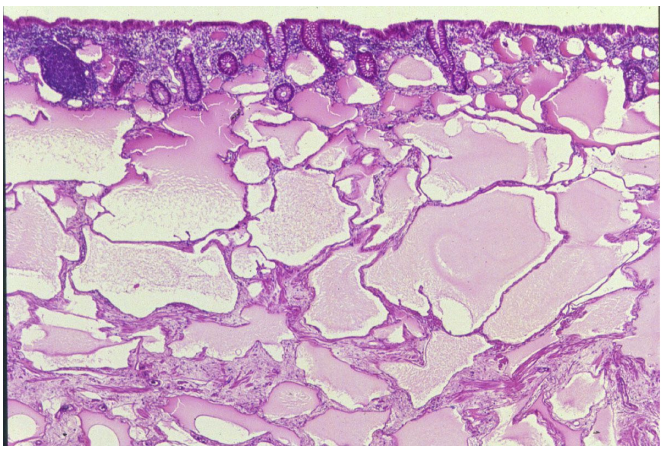

Fig. 6. Histopathological findings: The tumor was located mainly in the submucosal layer, and lymph vascular dilatation filled with lymph was noted in the mucosal to subserosal layers.

The tumor was initially reported by Chisholm et al.[1] in 1932, and the initial Japanese case was reported by Yoshitoshi et al.[2] in 1965. According to Matsuda et al.[3], 279 cases have been reported by 2001 in Japan. On search in Igakuchuozasshi between 1983 and 2006, 41 cases were treated with laparoscopic resection of lymphangioma, and laparoscopy-assisted surgery was performed in very rare case[4].

According to various reports, the age distribution was 1-83 years with a most frequent age of 60 years, and the incidence was higher in males at a sex ratio of 2-2. 5: 1 in Japan[3)5)6)].

The development site tends to be located in the right half of the colon[3].

Many cases were incidentally discovered. Abdominal pain was the most frequent symptom, followed by bloody stool, constipation, and diarrhea. Weight loss seen in this patient is not a typical finding, but protein-losing enteropathy associated with a large tumor has been reported[7], which may have resulted in weight loss. Fecal occult blood was also an atypical finding[8].

For the diagnosis, histological observation of lymph vascular dilatation and outgrowth in the submucosal layer and coverage by a single luminal endothelial cell layer is necessary. Histopathologically, Wegner et al. classified lymphangioma into: 1) simple lymphangioma, 2) cavernous lymphangioma, and 3) cystic lymphangioma. No malignant case has been reported. Although cases complicated by colorectal carcinoma have been reported, the complications incidentally occurred, and the causal relationship was unclear[10)11)].

On contrast enema, lymphangioma is generally noted as an intraluminal filling defect with a clear margin. The shape and size are altered by compression and double contrast radiography, but no marked change is noted on the mucosal surface.

On colonoscopy, the mucosa is ash-colored, compared to the surrounding, and tumors are light-permeable, transparent, and pedunculated. Bridging fold is not necessarily noted. The shape is altered by postural change and compression with forceps, and the fluctuation is called cushion sign[12]. The tumor in this patient was also transparent and pedunculated. The usefulness of endoscopic ultrasonography (EUS) for definite diagnosis has been reported[13)14)].

Regarding therapeutic policy, Karasawa et al.[15] reported that polypectomy as diagnostic treatment is applicable for 2-cm or smaller pedunculated and semipenduculated types, and polypectomy is applicable after lymph drainage by endoscopic puncture for larger and sessile-type tumors, because lymphangioma is benign, and no malignant case has been reported. Many cases of lymphangioma larger than 3 cm were surgically resected in Japan. Difficulty in application of polypectomy for large tumors, and large tumor-induced invagination and ileus requiring surgical resection have been reported[16].

Wang et al.[8] reported a case of laparoscopy-assisted

resection of colon lymphangioma, in which low-invasive surgery was performed because differentiation from malignant diseases was difficult due to a large tumor size. In our patient, the possibility of malignant tumor could not be ruled out because extramural advancement of the tumor was suspected on preoperative diagnosis, and thus, endoscopic treatment was difficult, and laparoscopy-assisted surgery was performed. Characteristic laparoscopic findings were noted: 1) Lymph vascular dilatation filled with lymph disseminated on the serosal surface, 2) light-permeability and translucency, 3) soft touch when compressed with forceps (laparoscopic cushion sign), and 4) similar lymph vascular dilatation in the surrounding adipose tissue.

To our knowledge, laparoscopy-assisted surgery was previously performed in very rare case in Japan, and the surgery was useful for the diagnosis and treatment. For cases of colon lymphangioma in which diagnosis or endoscopic treatment is difficult, laparoscopy-assisted surgery should be considered as one choice. (41 reports found by search for 'colon lymphangioma' in 1983-2006 Igakuchuozasshi, and references used these reports were referred)

CONCLUSION

We encountered a patient with colon lymphangioma and performed laparoscopy-assisted surgery. Since characteristic laparoscopic findings were noted, laparoscopic surgery is a useful for a diagnosis and the treatment of large lymphangioma of colon.

We reported the case with a literature review.

The abstract of this report was presented in the 55th Annual Meeting of the Japanese Society of Gastroenterological Surgery (July 2000, Miyazaki)

REFERENCES

1) Chisholm AJ, Hillkowitz P Lymphangioma of rectum. Am J Surg 1932; 17: 281-2,
2) Yoshitoshi Y, Oda T, Utsumi Y, et al. A case of lymphangioma of the ascending colon. Nihon Rinsho 1965; 23: 2264-7 (in Japanese)
3) Matsuda T, Matsutani T, Tsuchiya Y, et al, A clinical evaluation of the large intestine: a case presentation of the descending colon and a review of 279 Japanese cases. J Nippon Med Sch 2001; 68 : 262-5,
4) Sato Y, Okuda T, Takayama T,et al. A Case of Lymphangioma of the Colon with Overlapping Tubular Adenoma. Gastroenterol. Endosc. 2002; 44: 1180-85 (in Japanese)
5) Ohtani H, Fujimoto Y, Ikezoe M, et al. A case of lymphangioma of the large intestine with an anomalous shape –study of 137 cases reported in Japan-. Geka 1997; 59: 1135-8 (in Japanese)
6) Noto T, Yasuda S, Mukai M,et al. A Lymphangimoma of the Transvese Colon Removed by Endoscopic Polypectomy. Syoukakinaisikyo 1992; 4: 135-7 (in Japanese)
7) Zilko PJ, Laurence BH, Sheiner H, et al, Cystic lymphangioma of the colon causing proteinlosing enteropathy, Am J Dig Dis 1975; 20: 1076-80
8) Wang HS, Chen WS, Lin JK, et al, Diagnosis and Treatment of Cystic lymphangioma of the Ascending Colon by Laparoscopic-Assisted Surgery: A Case Report, Chin. Med. J. (Taipei) 1999; 62: 322-5
9) Wegner G, Lymphangiome, Arch. Klin. Chir. 1877;20:641
10) Morozumi K, Omoto T, Miyazaki H et al.Two cases of lymphmgioma of colon complicated colon cancer, Gastroenterol. Endoscopy 41: 1323-1329 1999 (in Japanese)
11) Miyamoto M, Nakajima K, Masuda K et al. A case of concomitant ascending colon lymphangioma and early rectal cancer. Hiroshima Igaku 1995; 48: 555-9 (in Japanese)
12) Kawamura N, Matsubara Y, Yahagi N et al. Soft and transparent submucosal tumor: Colon lymphangioma. Shokakinaishikyo 1998; 10: 208-209 (in Japanese)
13) Ohta A, Arai J, Sato K,et al. Endoscopic ultrasonography for diagnosis of lymphangioma of the colon. Prog. of Dig. Endoscopy 1998; 54: 94-5 (in Japanese)
14) Murakita H, Hirano S, Mizuno S,et al. Lymphangioma of the Colon Diagnosed by Endoscopic Ultrasonography. Gastroenterol Endosc 1991; 33: 2106-13 (in Japanese)
15) Karasawa H, Miura T, Makino T, et al. Two cases of the large intestine extracted by endoscopic polypectomy. Progress of digestive Endoscopy 1985; 26: 330-4 (in Japanese).
16) Kim TO, Lee JH, Kim GH, et al, Adult intussusception caused by cystic lymphangioma of the colon: a rare case report, World J Gastroenterol 2006; 12: 2130-