Lymphangioma of the colon : A case report and review of the Japanese literature

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Abstract

Intraabdominal lymphangiomas are rare, although these lesions can occur in the mesentery, omentum, retroperitoneum, or gastrointestinal tract wall. Here we report a case of lymphangioma of the transverse colon and review the other cases reported in the Japanese literature. Our patient presented with lower abdominal pain and barium enema revealed a filling defect in the transverse colon. Colonoscopy disclosed a submucosal tumor, which changed shape with alterati on of the patient's position and showed the cushion sign. This lesion was covered with normal-looking mucosa. A correct diagnosis was made from these findings. Knowledge of these endoscopic features may help physicians to provide appropriate diagnosis and treatment of colonic lymphangioma. (Acta gastroenterol. belg., **2000**, 36, **239-241**).

Key words : lymphangioma, large intestine, colonoscopy, non-epithelial tumor.

Introduction

Non-epithelial tumors of the colon are rare, and lymphangioma is one of these tumors. Since Chisholm *et al.* described this disorder for the first time in 1932, several cases have been reported in various countries (1-5). In Japan, with the development and widespread use of colonoscopy, this tumor has recently been reported more frequently (6-9). Here we describe a patient with lymphangioma of the colon and also review the relevant literature on other Japanese cases.

Case report

A 60-year-old woman was admitted to Higuchi Gastrointestinal Hospital in 1998 with a one-month history of intermittent lower abdominal pain and diarrhea. There was no history of weight loss or melena. Review of the family history and past medical history revealed no unusual features. Tests for anemia and hypoalbunemia were negative, as was the fecal occult blood test. Tumor markers showed no significant changes. Barium enema revealed an elevated lesion that was 3 cm in diameter with a smooth surface in the transverse colon. The lesion appeared to be a broad based submucosal tumor (Fig. 1). Colonoscopy subsequently showed that the tumor had a smooth surface, and was slightly yellowish and translucent compared with the adjacent normal mucosa, although it was covered with normal-looking mucosa. Its shape varied along with changes of the patient's position. A depression was easily made in the lesion by compression with biopsy forceps the cushion sign (Fig. 2). We diagnosed lymphangioma of the colon from these findings, making a differential diagnosis from among lipoma, hemangioma, and lymphangioma. Abdominal computed tomography (CT) showed a circumscribed, thin-walled cystic mass in the right upper quadrant.

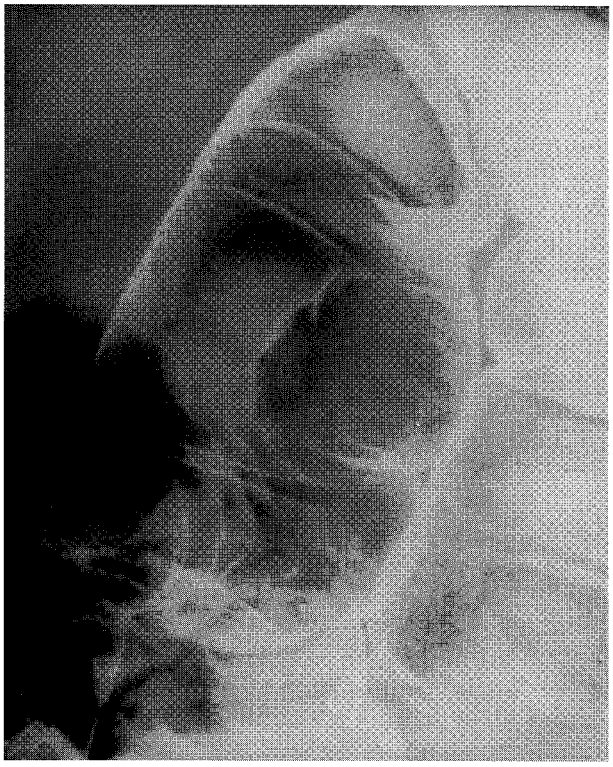

Fig. 1. — Barium enema reveals a smooth lesion in the transverse colon.

Poulos *et al.* recommended that endoscopic polypectomy to avoid the risk of obstruction for lesions smaller than 2 cm and surgical resection those for larger lesions because of the risk of colonic perforation (5). Based on their indications we performed the surgical resection of this lesion. Macroscopically, the resected tumor was 3 cm in diameter. It showed fluctuation and the fluid aspirated with a syringe was clear yellow. Microscopically, this tumor was confirmed to be a lymphangioma and the cyst fluid contained a great many lymphocytes (Fig. 3).

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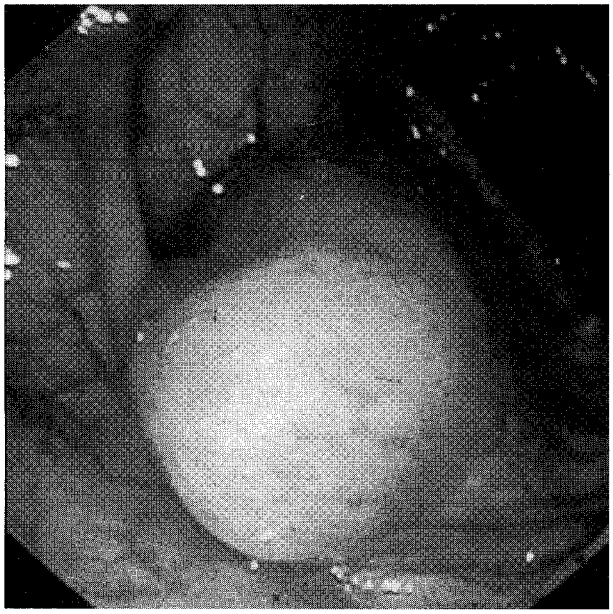

Fig. 2. — Colonoscopic view of the lymphangioma in the transverse colon.

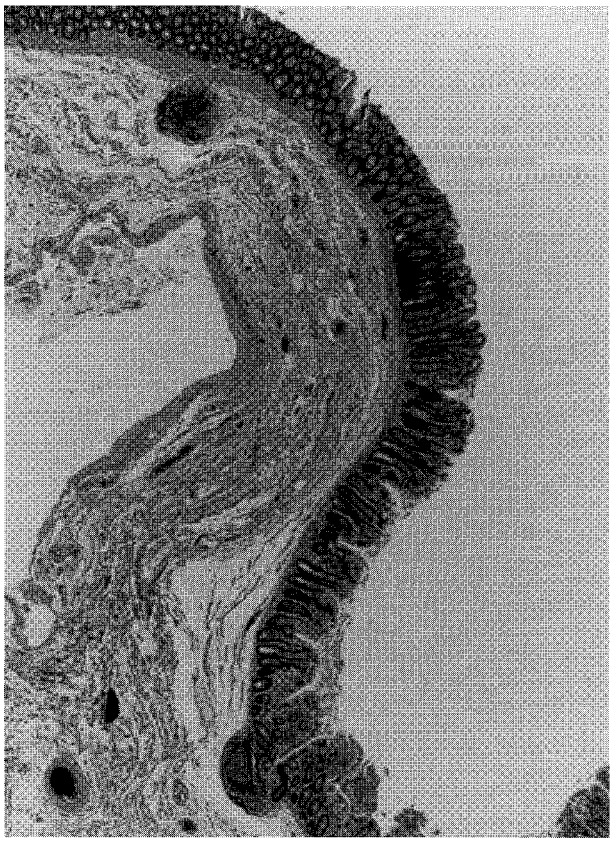

Fig. 3. — Microscopic examination of the tumor shows a lymphangioma with cystic spaces in the submucosa (hematoxylin and eosin, ×20).

Discussion

Lymphangioma is known to occur frequently in the face, chest, extremities, lips, tongue, eyelids and conjunctiva. Intraabdominal lymphangiomas are rare however, occurring in the mesentery, omentum, retroperitoneum, or gastrointestinal tract wall (10,11). Lymphangioma is the least common benign submucosal tumor of the colon. In 1943, Helwig did not find a colonic lymphangioma in a series of 1,460 autopsy cases (12). In 1970, Fleming *et al.* found only one among 453,708 patients who underwent roentgenographic examination of the gastrointestinal tract in a series of 1,437,767 cases (13). In Japan, the incidence of colonic lymphangioma appears to be higher than in America and Europe. In 1989, Okamoto *et al.* reported that there were 120 nonepithelial colonic tumors (2.8%), including 20 lymphangiomas (0.47%), among 4,211 polpoid lesions removed by colonoscopic polypectomy (14). Hara *et al.* reported six cases of lymphangioma in a review of 1130 autopsy cases (15). Our literature search showed that 128 cases, including the present one, have been reported in Japan. The mean age (±SD) was 51.2±13.7 years and there were 90 men (72%) and 33 women (28%), (5 cases had no description of gender). There were 44 lesions (36%) in the transverse colon, 36 cases (29%) in the ascending colon, 16 cases (13%) in the cecum, 14 cases (11%) in the descending colon, 8 cases (7%) in the sigmoid colon and 6 cases (4%) in the rectum. The lymphangioma was under 2 cm in diameter in 57 patients (58%), while it was equal to or larger than 2 cm in 42 patients (42%), (29 cases had no description of size). Signs and symptoms associated with lymphangioma were abdominal pain (42%), melena (16%), diarrhea or constipation (19%), and others (23%).

This tumor was also found incidentally during routine endoscopic and radiographic studies of the colon. Some reported cases were complicated by stenosis or intussusception (1,16).

Endoscopic findings were a round and smooth appearance, a broad base, a pale blue or translucent color, easy a change in the shape with postural change, and a positive cushion sign when compressed with forceps (17). Radiographic examination revealed oval to round radiolucent defects with sharply defined borders, and a tendency for changes in lesion shape and size on compression or double contrast radiography (2). The diagnosis of colonic lymphangioma has usually been made by endoscopy, radiography, or both. However, the preoperative diagnosis rate for this disease was only 27.3% in Japanese cases. Thus, the physician must have knowledge about these characteristic endoscopic features in order to make an appropriate diagnosis. Recently, some investigators reported the possibility of diagnosing lymphangioma with computed tomography (CT) or endoscopic ultrasonography (9). In our case, we could easily diagnose lymphangioma by colonoscopy, which is a simple and widely used procedure.

Asymptomatic lymphangioma of the colon can be left alone, but symptomatic lesions or those which are difficult to diagnose require treatment. Surgical resection is recommended for lesions larger than 2 cm, because of the risk of colonic perforation. For smaller lesions, simple aspiration, polypectomy, and injection with ethanol are now advocated (7,9). Regarding the treatment of the 128 cases reported in Japan, 59 (50%) underwent surgical resection, 59 (50%) underwent endoscopic polypectomy, and 10 cases had no description of treatment. Recently, treatment by polypectomy has been increasing, but we performed surgical resection because of the size of our patient's lesion.

Our experience suggests that clinicians should consider the possibility of lymphangioma of the colon on endoscopic examination based on the characteristic findings of a change in shape with patient position and a positive cushion sign.

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