J Korean Med Sci 2001; 16: 229-32
ISSN 1011-8934

Gastric Lymphangioma

Gastric lymphangioma is a rare benign gastric tumor composed of unilocular or multilocular lymphatic spaces. On gastrofiberscopy a submucosal tumor covered with smooth transparent normal mucosa is revealed in the stomach with or without a stalk. Endoscopic ultrasonography has become an indispensable tool for differentiating these gastric tumors. Treatment of lymphangioma depends on its size, location, and presence of complications. Endoscopic resection is safe and easy and plays an important role in confirming the diagnosis and treatment of the tumors especially of small-sized ones. We report a case of gastric lymphangioma in a 68-yr-old woman who presented with nausea and vague epigastric discomfort for two months. She was diagnosed by gastrofiberscopy with endoscopic ultrasonography and treated successfully with endoscopic resection by strip biopsy method.

Key Words: *Lymphangioma; Stomach; Endosonography; Gastroscopy*

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Received: 14 January 2000
Accepted: 23 June 2000

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INTRODUCTION

Lymphangioma is a benign lymphatic vascular tumor which is commonly found in the head, neck, axilla, skin and occasionally in the mesentery or gastrointestinal tract (1). Among them, gastric lymphangioma is very rare and has been reported in less than 100 cases worldwide since 1953 including several cases reported in Korea (2-8).

Endoscopic examination has been a popular method of diagnosing upper gastrointestinal disease. More specifically, endoscopic ultrasonography (EUS) is the most valuable method for the differential diagnosis of submucosal tumors of the stomach (9). Confirmatory diagnosis and management of gastric submucosal tumor are possible by EUS and endoscopic resection technique respectively (8, 9).

We report a case of gastric lymphangioma which was diagnosed by EUS and treated with endoscopic resection.

CASE REPORT

A 68-yr-old woman previously healthy was admitted to our hospital in 1999 due to nausea and vague epigastric discomfort for two months. These symptoms were associated with intake of food. Medical history of her family members revealed her brother who had been diagnosed as stomach cancer.

On admission, her vital signs were stable. Physical examination findings were unremarkable: it did not reveal any palpable lymphadenopathy on the neck, abdominal mass, ascites, or hepatosplenomegaly. Laboratory findings such as complete blood count and blood chemistry were normal.

Gastrofiberscopic examination showed a well-demarcated cystic mass, about $25 \times 20 \times 10$ mm in size, in the anterior wall of the distal body. The surface of tumor was smooth and mildly transparent in nature, and the tumor was soft enough to be compressed with biopsy forceps (Fig. 1).

Endoscopic ultrasonography with Olympus EU-M30 main unit and conventional EUS (GF-UM200) was performed. It showed a cystic mass located in the submucosal layer, which was anechoic in nature with several inner septations (Fig. 2).

We removed the tumor by endoscopic resection with strip biopsy method (Fig. 1). First we injected epinephrine-hypertonic saline solution diluted at 1:10,000 into the margin of the tumor to separate the tumor from the muscular layer. Resection was performed using Olympus snare and Olympus UES-20 diathermy. Complications such as abdominal pain, bleeding, or perforation were

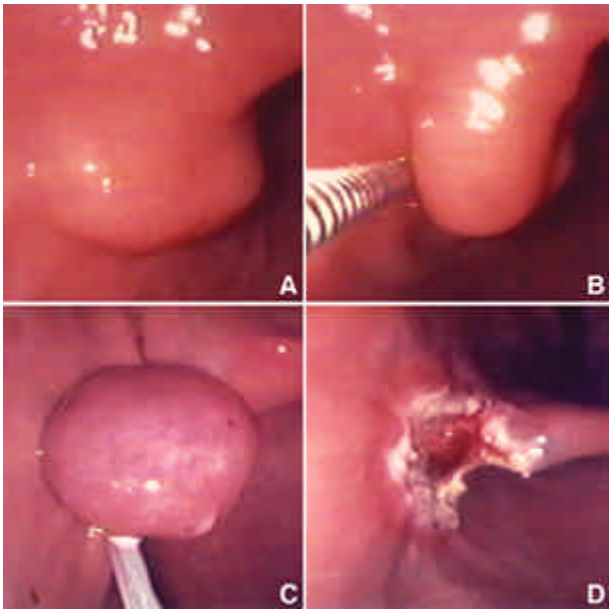

Fig. 1. Endoscopic resection with strip biopsy method. (A) Upper gastrointestinal endoscopy shows a well-demarcated mass covered with smooth normal gastric mucosa in the anterior wall of the distal body. (B) Epinephrine-hypertonic saline solution was injected into the margin of the tumor. (C) The tumor was resected using a electric snare. (D) The remaining ulcer shows an even base with clear resection margin without significant hemorrhage.

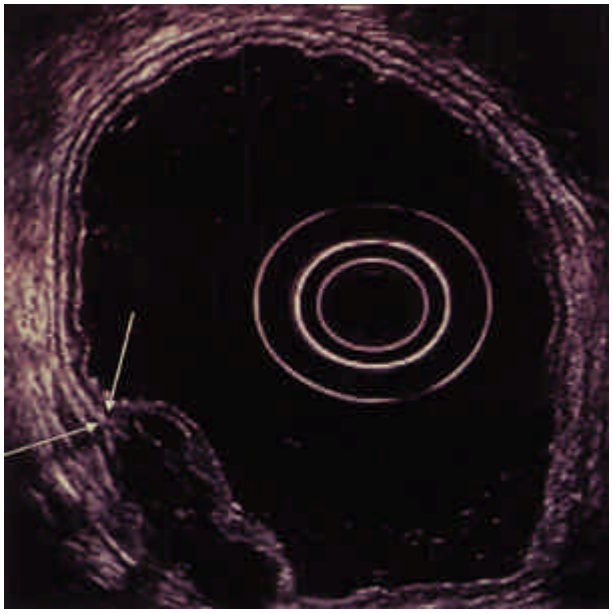

Fig. 2. Endoscopic ultrasonography shows a cystic anechoic mass with multiple inner septations located in the third hyperechoic layer. The arrows indicate two muscular hypoechoic layers which consist of the muscularis mucosa and muscularis propria.

absent.

Microscopic examination of the removed tumor disclosed a dilated cystic spaces lined with flattened endothelial cells in the submucosa. The cystic spaces contained lymphatic fluid without red blood cells. Focal areas of lymphocyte aggregation were noted. The adjacent layers of mucosa and muscularis mucosa remained relatively normal (Fig. 3).

The patient was discharged without any complication and has been followed up uneventfully.

DISCUSSION

Lymphangioma is a benign tumor which is composed of a single or multiple thin-walled lymphatic spaces, fibrous tissue, aggregates of lymphoid cells and lymphatic fluid. It occurs mostly in the head, neck, axilla, trunk and rarely in the internal visceral organs (1). Among the intra-abdominal lymphangiomas, the mesentery is the most frequently involved site and other intra-abdominal organs such as the peritoneum, omentum, retroperitoneum, liver, gallbladder, spleen, and gastrointestinal tract can be involved (10, 11). Gastrointestinal lymphangioma can occur anywhere along the gastrointestinal tract, being the small bowel the most common site of origin (12-15). Since the first report in 1953 (16), less than 100 cases of gastric lymphangioma have been reported worldwide including Korea (2-8).

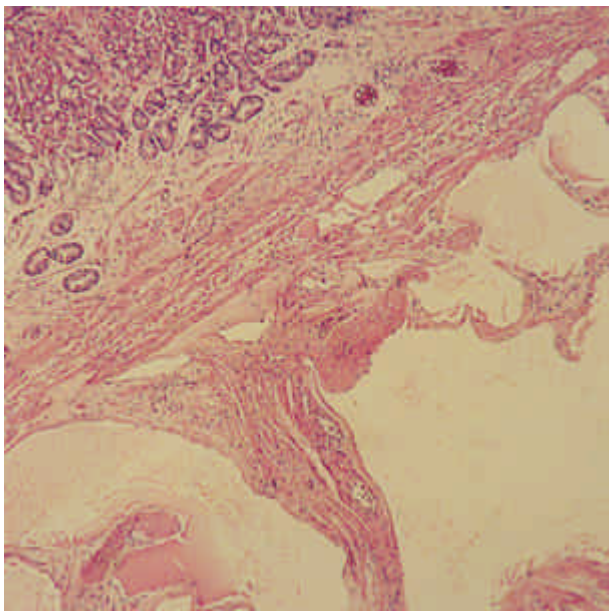

Fig. 3. Pathologic finding of the resected specimen reveals multiple cystic dilated lymphatic vessels, which contain eosinophilic fluid without red blood cells in the submucosa (H&E, ×40).

Yamaguchi et al. (5) reviewed gastric lymphangiomas reported in the literatures along with their own case. Age of the patients ranged from 5 to 79 yr with a mean age of 47 yr. Seventeen were male and 15 were female.

Most patients with gastric lymphangioma are asymptomatic or experience vague upper abdominal discomfort depending on their location, size and degree of obstruction. Other presenting symptoms include abdominal pain, nausea, dyspepsia, hemorrhage, obstruction and palpable mass (2-8). In our case, the patient complained of nausea and vague epigastric discomfort without other symptoms such as hemorrhage or obstruction.

Gastric lymphangioma can be diagnosed as a submucosal tumor by gastrofiberscopy. The typical endoscopic findings thereof are sharply demarcated oval to round-shaped and yellowish-white to tan-colored nodular mass with or without a stalk. The surface is smooth and transparent, often with white specks. Lymphangioma is readily compressed by biopsy forcep and the shape is changeable by peristalsis. Deeper biopsy can release milky fluid (2-7).

Most of gastric lymphangiomas are found as a single tumor located in the submucosal layer of the stomach (5). EUS has recently become a standard tool in assessing gastric submucosal tumors. The characteristic EUS findings of lymphangioma are an anechoic lesion in the submucosal layer with multiple inner septations in the cyst (8). By using EUS, gastric lymphangioma can be differentiated from submucosal tumors such as lipoma, hemangioma, ectopic pancreas, carcinoid tumor, leiomyoma, benign submucosal cyst, solitary submucosal varix, early epithelial polyp, and others.

Most lymphangiomas in the gastrointestinal tract have a benign course. However, lymphangiomas combined with gastric adenocarcinomas have been reported, and whether this is an incidental finding or a true association is still in doubt (2, 5, 8).

Treatment of gastric lymphangioma often depends on its size, location, and presence of complications. Surgical procedure was accepted as the only a method to treat large symptomatic lymphangiomas and some investigators have suggested that small asymptomatic lymphangiomas need only to be followed-up. Recently several endoscopic managements such as endoscopic aspiration, endoscopic excision, endoscopic injection with sclerosing agent and endoscopic resection have been increasingly in use and have become the treatment of choice rather than surgical operation especially for small-sized tumors. Since conventional endoscopic forcep biopsy can not yield a final diagnosis of submucosal tumors, endoscopic resection, which is safe and easy, plays an important role in confirming diagnosis and treating lymphangioma while avoiding surgical risks (2, 3, 17, 18).

The size of gastric lymphangiomas in reported cases varies, ranging from 10 to 280 mm (2-8). Kajiyama et al. (18) reported that the success rate of endoscopic resection varied with tumor size. In cases of gastric submucosal lesions less than 2 cm in diameter, the eradication rate was about 85-94%. In our case, the size of tumor was about $25 \times 20 \times 10$ mm and the tumor was successfully resected with EUS without any complications.

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