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Giant Mesenteric Cystic Lymphangioma Originating from the Lesser Omentum in the Abdominal Cavity

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A 48 year old woman was diagnosed with a huge cystic mass in her abdominal cavity. She complained of significant abdominal discomfort due to the mass. The abdominal computed tomography revealed a giant multi-lobulated mass, measuring 26×12 cm in size, adjacent to the lesser curvature of the stomach. In the operation field, the mass was found to originate from the lesser omentum, including the right and left gastric vessels and the vagus nerves, and to invade the lesser curvature of the stomach. For curative resection, distal subtotal gastrectomy with mass excision followed by gastroduodenostomy were performed. This mass was pathologically diagnosed to be a mesenteric cystic lymphangioma; in fact, the largest ever reported. The patient had no complications during the postoperative period and was discharged from the hospital on the seventh day after surgery.

Key Words: Lymphangioma; Gastrectomy; Omentum

Introduction

While cystic lymphangiomas are often diagnosed in the head and neck and the axilla of infants, this tumor is notably rare in the abdominal cavity.(1,2) Because of the low incidence and vague symptoms of lymphangiomas in the abdominal cavity, they are often unexpectedly diagnosed as large size multi-cystic tumors in the operative field of other diseases.(3) Most lymphangiomas in the abdominal cavity originate from the mesentery in which most lymphatic channels are included. Therefore, the lymphangiomas in the abdominal cavity were previously called "mesenteric cystic lymphangiomas (MCLs)". To date, various sizes and locations of MCLs have been reported, and intestinal resection is rarely required to pathologically diagnose and cure them.(4,5)

The greater and lesser omentums, which are attached in the stomach, are typical sites of MCLs in the intraabdominal cavity. MCLs near the stomach can be found as submucosal tumors in the gastrofiberscope, and large lymphangiomas can induce mass effect symptoms. Although most MCLs in the omentum can be removed by omental excision, gastrectomy may be required, depending on the tumors' locations and characteristics. To the best our knowledge, three cases of MCL from the omentum near the stomach have been reported, but the sizes of these tumors were below 20 cm.

Recently, we diagnosed a giant MCL of over 20 cm in size that originated from the lesser omentum in the abdominal cavity. This tumor was completely resected by gastrectomy.

Case Report

A 48 year old woman who was diagnosed with a huge intraabdominal cystic mass was transferred to our department. She had been diagnosed with breast cancer two months earlier and had un-

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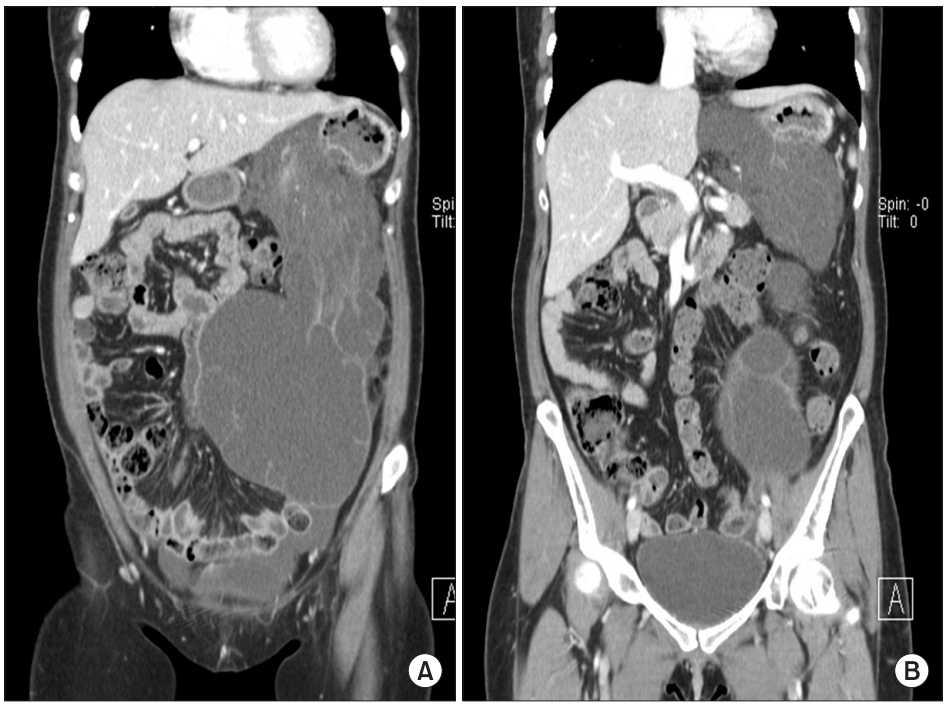

Fig. 1. Findings of the computed tomography analysis. (A) A multi-lobulated cystic mass was observed to extend into the pelvic cavity. (B) This mass originated from the lesser omentum, including the truncal vagus.

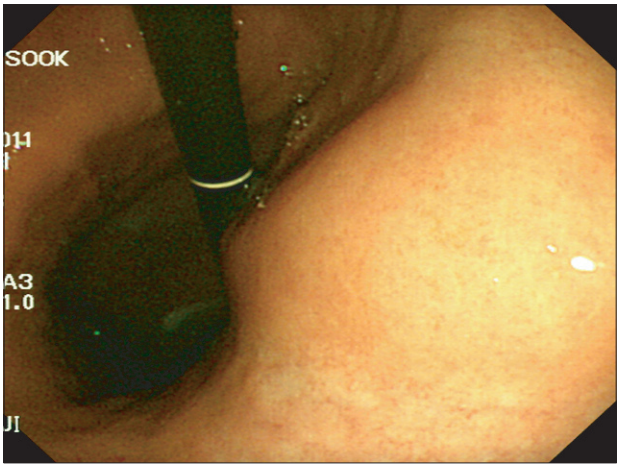

Fig. 2. Gastrofiberscopic findings showing the narrowing of the stomach lumen by the extrinsic compression of the lesser curvature. However, the mucosa was intact.

dergone partial mastectomy with sentinel lymph node biopsy. After surgery, adjuvant chemotherapy of three cycles was administered. She had recently developed intermittent abdominal pain. Under physical examination, a diffuse soft mass was palpable with mild tenderness, but the margin of the mass was not clear. Abdominal computed tomography (CT) for evaluating the abdominal pain and mass revealed a giant multi-lobulated mass, measuring 26×12 cm in size, which was adjacent to the lesser curvature of the stomach (Fig. 1). However, the origin of this mass was not determined by CT. The gastrofiberscopic findings revealed a large bulging lesion with intact overlying mucosa on the posterior wall and lesser curvature side of the stomach (Fig. 2). All blood laboratory data values were within normal ranges. Based on the physical examination and the imaging study, we decided to perform exploratory laparotomy after obtaining informed consent from the patient.

After the upper midline incision, a large multi-cystic mass originating from the lesser omentum of the stomach was detected. A portion of the mass adhered to the omentum; this adhesion was easily dissected, but the lesser omentum, including the right and left gastric vessels and the vagus nerves, were fully involved in the mass. The mass appeared to invade the lesser curvature of the gastric antrum. We decided to perform distal gastrectomy with mass excision for curative resection.

First, the gastrohepatic and gastrocolic ligaments were dissected to mobilize the stomach and the mass, and the right gastric vessels were resected. The right gastroepiploic vessels were resected, and the first portion of the duodenum was resected. The posterior wall of the stomach was exposed, and the left gastric vessels were subsequently resected. Because the mass involved the trunk of both vagus nerves near the abdominal esophagogastric junction, truncal vagotomy and dissection of the lesser omentum from the upper portion of the lesser curvature were performed. Finally, the distal portion of the stomach was resected, and the mass with the distal stomach was extracted without any lymphatic leakage. The gastroduodenostomy was performed with a 29 mm circular stapler.

The specimen was 22×10×5 cm in size, and its cross-section

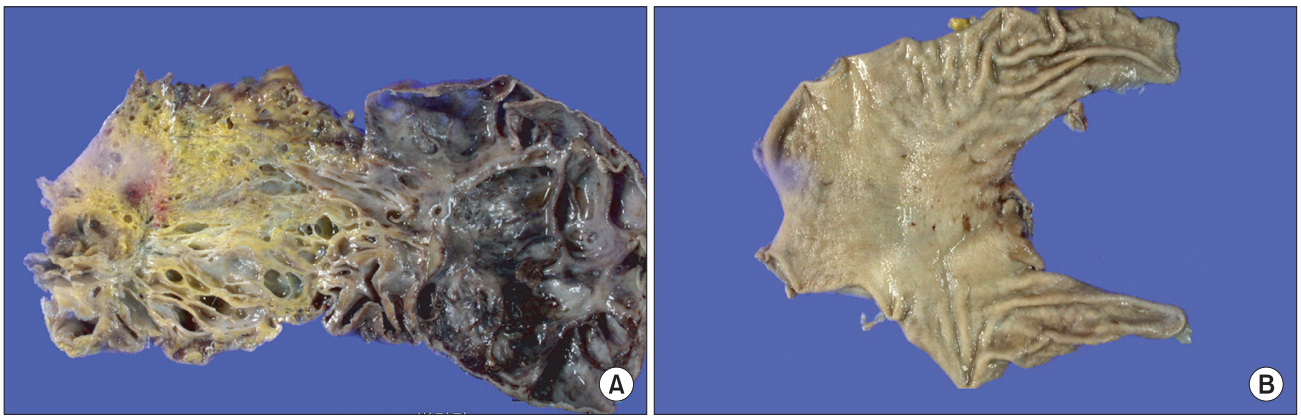

Fig. 3. Photo of the macrosopic findings of the specimen. (A) Transected tumor showing the multilobulated cystic tumor, including the partially solid portion. (B) The stomach specimen showed normal mucosa after opening of the greater curvature.

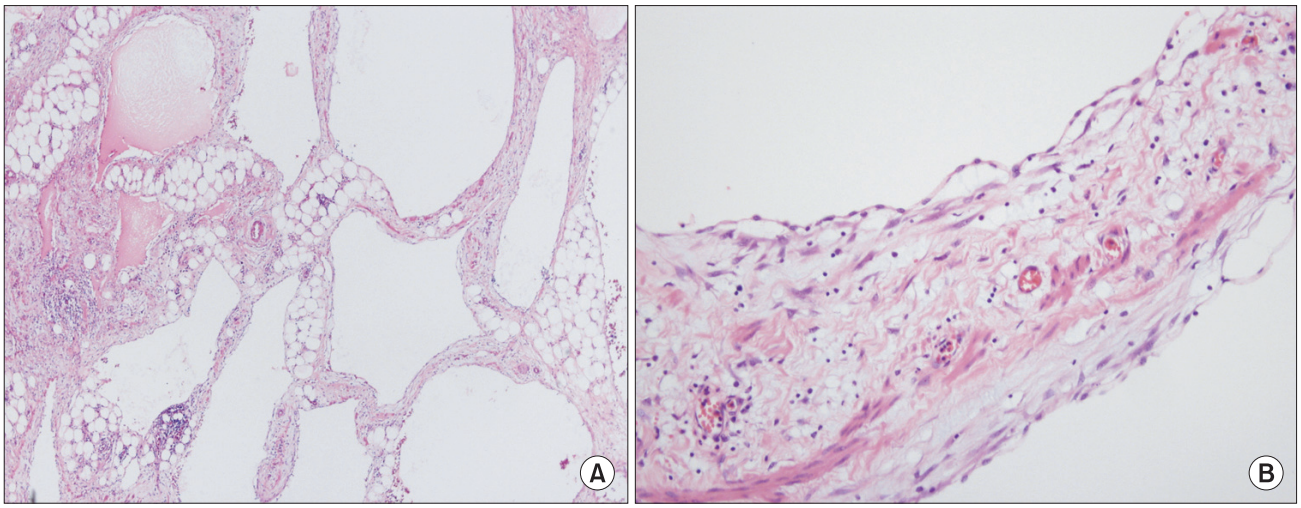

Fig. 4. Photo of the microscopic finding of the specimen. (A) The tumor was composed with multiple cysts of various size (Hematoxylin-Eosin Stain, ×40). (B) Each cyst was lined with the flat endothelial cells, and filled proteinaceous material. Several inflammatory cells were observed in the intercystic space (Hematoxylin-Eosin Stain, ×200).

presented as a multilobular cyst (Fig. 3A). The tumor was slightly attached to the lesser curvature of the stomach, but the mucosa of the stomach was intact (Fig. 3B). In the microscopical finding, tumor was composed with the multiple cysts which were various sizes, and lined with the endothelial cells (Fig. 4).

The patient had no complications during the postoperative period, and oral intake began on postoperative day 3. She was discharged from the hospital on the seventh day after surgery. Additional adjuvant treatment for breast cancer was started again, and the patient had no complaints related to the gastrointestinal tract 6 months after surgery.

Discussion

Lymphangiomas are benign tumors, but the cause of this disease is not clear. Congenital or traumatic failure of the lymphatic system are the most common causes.(6) The common sites for this tumor are the neck (75%) and axilla (20%), but occurrence in other areas, such as the mediastinum and the abdominal cavity, are rarely seen as well. Although cystic lymphangiomas in the abdominal cavity were previously considered mesenteric cysts, the histological difference between lymphangiomas and mesenteric cysts was recently established. In contrast with mesenteric cysts, which originate from mesothelial tissue, lymphangiomas are composed of alternating lymphoid tissue, lymphatic space and foam cells.(3) As most lymphangiomas in the abdominal cavity originate from the mesentery and form multi-lobular cysts, we refer to them as MCLs.

Although MCLs occur at all ages, over 60% of patients are diagnosed before 15th year of life.(7) However, it has been reported that the prevalence of this tumor is as rare as 1 in 20,000 to 250,000

people.(3,8) Cystic lymphangiomas in the neck and axilla present as palpable masses at birth, and at least 90% become symptomatic before the second year of life.(7) However, most patients with MCLs are initially asymptomatic. Unless this tumor is accidentally diagnosed during laparotomy, diagnosis is most often not made until the size of the tumor becomes large enough to cause symptoms, including abdominal discomfort and partial intestinal obstruction.(9) Rarely, MCLs can lead to acute complications, including severe abdominal pain due to rapid growth, traumatic rupture, intraabdominal bleeding and complete obstruction. These patients require emergency laparotomy.(10,11) The patient in our case was not diagnosed until the tumor size increased to over 20 cm, despite the fact that she had been regularly followed after mastectomy for breast cancer. Fortunately, her tumor was removed by surgery before it caused severe complications.

The treatment of choice for MCL is surgical resection. In spite of this tumor's benign nature, it tends to invade into adjacent structures and to recur after curative resection.(3) Therefore, the principle of surgical resection for MCL is to remove the total lesion, including the entire mass and the invaded organs, such as the bowel, the pancreas or the spleen.(12) In our case, the tumor included the entire lesser omentum, both vagus nerves, and the right and left gastric vessels, and it invaded the lesser curvature of the stomach. In order to complete the total resection of this tumor, we performed distal gastrectomy. Recently, laparoscopic removal of MCLs has been shown to be feasible.(13) This technique has several advantages over conventional laparotomy, including minimal trauma, reduced pain, and early recovery after surgery. However, with the giant tumor in our case, it was impossible to dissect by laparoscope because the tumor filled a large portion of the abdominal cavity.

Losanoff et al.(14) reported in their review article that MCLs can be classified into four types. Type I, the pedicled type, can cause torsion and necrosis or rapidly enlarge. Type II, the sessile type, is located within the mesentery of the bowel. Type III, the retroperitoneal type, involves the retroperitoneal structures, such as the mesenteric root, the aorta and the vena cava. The type IV multicentric type extensively involves the intraabdominal and retroperitoneal organs. This classification can establish the treatment strategy for mesenteric cystic lymphangiomas. Most tumors of type I can be removed by excision, and type II tumors may require combined resection of the bowel involving the mesentery. For tumors of type III and type IV, palliative treatment methods can be used, including partial resection and marsupialization.(15) The patient in this case report presented a tumor of type II, suggesting that the surgical procedure should be resection of the bowel. However, the stomach has a significant blood supply, due to its curvature.

To date, three cases of MCL in the stomach have been reported by case report and case series.(4,5,13) The sizes of these tumors were 5, 9 and 20 cm, respectively. While the 5 and 9 cm tumors were resected by omental excision without gastrectomy, the 20 cm tumor required gastrectomy. Omental excision alone is not appropriate for tumors measuring over 20 cm, because of the high incidence of the involvement of major vessels and the vagus nerves and invasion into the stomach parenchyma. Gastrectomy for resection of this type of tumor is required.

In conclusion, we report a giant MCL that originated from the lesser omentum that was resected with distal gastrectomy for curative treatment. This MCL is the largest one that has been reported to date. This giant MCL from the mesentery near the stomach required the gastrectomy to resection completely.

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