An esophageal duplication cyst is a cyst originating in the foregut. These cysts are relatively rare, accounting for only 0.9 to 2.5% of mediasternal tumors. A patient presented with a duplication cyst from the abdomen, which had perforated into the esophageal lumen with a consequent abscessation. This cyst was surgically resected. The patient was a 57-year-old man and he presented at a nearby hospital with epigastric pain in November 2006. An examination revealed a mass perforating into the esophageal lumen with a hemorrhage directly above the esophagogastric junction. Although conservative treatment was administered for 4 months, the mass was associated with abscessation and progressed into an intractable condition. Therefore, the patient was referred for surgical treatment. An examination revealed a nearly semicircular submucosal tumor situated just superior to the esophagogastric junction and persistent draining was also noted from its central recess. Abdominal computed tomography, magnetic resonance imaging scans and echography demonstrated a cystic lesion, 50 mm in diameter, containing fluid, walled with a thick septum having an irregular luminal surface and abutting on the wall of the abdominal esophagus. A surgical resection was indicated for this condition because the possibility of malignancy could not be ruled out based on the diagnostic imaging results. A laparotomy with a lower esophagectomy and fundusectomy were performed in combination with resection by jejunal interposition. After the operation the patient’s condition was favorable and he is now being followed on an outpatient basis. The histopathological diagnosis of the present case was a duplication cyst associated with the microscopic features of inflammation.

**Keywords:** Esophageal cyst, Duplication cyst

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recess (Fig. 2).

An abdominal ultrasound study showed a cystic lesion, measuring 40×52×42 mm, containing two-layered fluid and abutting on the anterior wall of the lower esophagus. The wall of the cyst was partially thickened with an irregular luminal surface (Fig. 3).

Thoracoabdominal computed tomography demonstrated an internally low-density cystic lesion, measuring 47×51×40 mm, in the anterior wall of the abdominal esophagus. The cyst was thickly walled, 7 mm in maximum thickness and the luminal surface was papillately irregular in part with a pronounced contrast effect. There were sporadic lymph nodes about 5 mm in diameter in the region around the cardia (Fig. 4).

Chest magnetic resonance imaging revealed a sharply demarcated cystic lesion, 53×49×42 mm in size, adjacent to the anterior wall of the abdominal esophagus. The wall of the cyst showed high-intensity areas on the T1-weighted scan and low-intensity areas on the T2-weighted image. The luminal surface of the wall showed an irregular contour in part, indicative of an elevated lesion. The cystic contents were visualized as low-intensity and high-intensity areas on the T1- and T2-weighted images, respectively. Several lymph nodes a few millimeters in diameter were noted in the pericystic region (Fig. 5).

Fig. 1 An esophageal radiography revealed an elevated lesion with relatively smooth margins in the lower to abdominal esophagus (arrows), protruding into the esophageal lumen.

Fig. 2 An endoscopy disclosed a nearly semicircular submucosal tumor in the right, anterior wall just above the esophagogastric junction and persistent white-colored fluid with a foul smell draining from its central recess (arrow).

Fig. 3 An abdominal ultrasound study showed a cystic lesion, containing two-layered fluid and abutting on the anterior wall of the lower esophagus. The wall of the cyst was partially thickened with an irregular luminal surface (arrows).

Fig. 4 Abdominal computed tomography demonstrated an internally low-density cystic lesion, in the anterior wall of the abdominal esophagus. The cyst was thickly walled and the luminal surface was papillately irregular in part with a pronounced contrast effect (arrows).

Fig. 5 Magnetic resonance imaging revealed a sharply demarcated cystic lesion, adjacent to the anterior wall of the abdominal esophagus (arrows). The wall of the cyst showed high-intensity areas on the T1-weighted scan (A) and low-intensity areas on the T2-weighted image (B).
The results of the pus cultures showed *Haemophilus parainfluenzae* (3+), *Neisseria sp.* (2+), *non-hemolytic streptococci* (2+) and α-*hemolytic streptococci* (2+). The cytological findings showed no malignant cells besides a finding indicative of Class II inflammation.

Therefore, since a gastrointestinal stromal tumor or malignant tumor could not be ruled out based on the diagnostic imaging results and the lack of response to the 4-months of conservative treatment, it was determined that surgical intervention was therefore indicated for this case. An operation was performed by laparotomy in May 2007. The surgical en-block resection of the tumor required a lower esophagectomy and a fundusectomy.

The laparotomy, via a midline incision in the upper part of the abdomen, revealed a smooth-surfaced, elastic, soft mass measuring 50 mm in diameter and adjoining the right to anterior aspects of the abdominal esophagus. The mass was found to have grown in the lesser omentum without adhesions to the surrounding structures yet it was fixed onto the lower esophagus, cardia and the upper part of the lesser curvature of the stomach. A lower esophagectomy and a fundusectomy were performed with an ensuing reconstruction by jejunal interposition. Since the mass was confined to within the subphrenic peritoneal cavity, it was possible to perform an en-block resection of the mass utilizing abdominal maneuvers alone.

The resected tissue specimen showed that the mass was a thick-walled cystic lesion containing white pus that had a foul smell. There were fistulations in the esophagus and the cyst. No obvious neoplastic lesion was noted on the luminal surface of the cyst (Fig. 6).

Histopathology revealed a duplication cyst with a wall containing the mucosa showing a pronounced chronic inflammatory cell infiltration, mainly by plasma cells and two well-grown smooth muscle layers (Fig. 7). There was no microscopic evidence of bronchogenic cyst-like cartilaginous tissue or malignancy. The lymph nodes in the surrounding area showed hyperplasia of the lymph follicles with no indications of malignancy.

The patient’s postoperative course was uneventful and he was discharged after significant improvement on the 16th hospital day. He is now well, remaining free from symptoms of reflux esophagitis, with a satisfactory eating status and a well-maintained quality of life one year following the surgical treatment.

**Discussion**

An esophageal duplication cyst is a congenital cyst of foregut origin that results from duplicate luminal formation while expanding the lumen due to vacuolization of the esophageal epithelium during the development of this organ. Histologically, the lumen of the cyst is covered with stratified squamous epithelium or embryonal pseudostratified epithelium resembling the esophageal epithelium and is characterized by being lined with two smooth muscle layers. It is a relatively rare disease which accounts for from 0.9 to 2.5% of mediasternal tumors.

Gastrointestinal duplication cysts have been reported to occur in such regions as the esophagus, stomach and duodenum. The most common affected site is the right side and the lower part of esophagus and the site in the present case coincided with this loci.

The clinical manifestations associated with a duplication cyst include esophageal pressure symptoms such as dysphagia, as well as precordial discomfort and arrhythm-
mia, while about 70% of cases are asymptomatic. In the case presented herein, the patient presented with fairly acute epigastric pain and a buckache possibly associated with the perforation of the cyst.

There have been some cases of this disorder in which there was perforation into the mediastinum besides those involving perforation into the esophagus. In the present case, perforation into the esophagus with hemorrhage followed by a retrograde infection due to closure of the mucosal aspect, resulting in abscessation. Will reported that endoscopic fenestration and drainage of the cyst provided a cure in one case; whereas in the present case, relapses were associated with spontaneous closure of an incised cyst after repeated interventions by endoscopic incision and drainage, thus leading to the conclusion that such conservative treatment was ineffective in this patient.

Tani performed a resection of the cyst after long-term follow-up with a diagnosis of leiomyoma of the esophagus, while Kuwajima reported their cases involved infection and abscessation which eventually led to the resection of the cyst as in the present case. There have also been reports of cases requiring a resection because of aggravation due to concurrent primary squamous cell carcinoma or a metastatic adenocarcinoma in the cyst. It would be advisable to resect the lesion early rather than to gratuitously follow the clinical progress over an extended period in cases of this disorder, taking into account the difficulty in making a definite diagnosis based on imaging data and possible malignant complications.

As for the surgical procedures, although the resection of the cyst alone by conserving the esophagus was reportedly achieved in one case, an en-block resection encompassing the esophagus was undertaken in the present case because 1) a possibility of a malignant neoplasm including the gastrointestinal stromal tumor could not be ruled out according to the preoperative diagnostic imaging data, 2) the cyst had perforated into the esophagus with fistulations between the esophageal lumen and the cyst and 3) there was also evidence of abscessation.

In the present case, the surgical reconstruction consisted of a lower esophagectomy combined with a fundusectomy and the subsequent jejunal interposition between the esophageal stump and the remaining stomach, rather than a direct esophago-gastric anastomosis, in consideration of the patient’s non-elderly age of 57 years and the postoperative quality of life. After the operation, the patient has remained free from symptoms of reflux esophagitis or subjective symptoms of depressed food propulsive function such as a heavy feeling of the stomach, with a satisfactory eating status and a well-maintained quality of life, thus indicating the technical success of the surgical procedures performed on this patient.

References