Submucosal Lymphatic Cyst of the Stomach

—A Case Report—

Young Jun Kim¹, Kyung Sup Chung¹, Jae Bock Chung¹, Sang In Lee¹, In Suh Park¹, Heung Jae Choi¹, Kyung Sik Lee³, Nam Hoon Cho¹ and Chan Il Park³

A submucosal lymphatic cyst is a thin-walled cyst, lined by flattened lymphatic endothelium, containing thin serous fluid. It rarely causes clinical symptoms, and it is incidentally discovered during fiberoptic panendoscopy or radiologic study in most cases. It is an extremely rare benign tumor of the stomach; however, a submucosal lymphatic cyst should be considered if a pliable and benign submucosal lesion is detected during fiberoptic panendoscopy. We report a case of submucosal lymphatic cyst of the stomach which showed a typical clinical picture. This report is the first case of submucosal lymphatic cyst of the stomach in Korea to the best of our knowledge.

Key Words: Submucosal lymphatic cyst of the stomach

Intramural lesions of the gastrointestinal tract are relatively uncommon but may occur at any site throughout its length. The lesions may be derived from any one of the mesodermal elements that compose the gastrointestinal tract wall or from the ectodermally derived nerves in the wall. Malignant neoplasms that are metastatic to the bowel wall from distant organs or from other parts of the gastrointestinal tract may also occur as intramural lesions (Fleming and Carlson 1970).

The common diseases which show an intramural nodule on fiberoptic panendoscopy or upper gastrointestinal series are leiomyoma, aberrant pancreas, intramural hematoma, isolated varix, dermoid cyst, and lipoma; these benign diseases should be differentiated from the submucosal lymphatic cyst (Fleming and Carlson 1970; Palmer 1951). A submucosal lymphatic cyst is a cystic dilatation of submucosal lymphatic vessels, and has a benign course. It rarely causes clinical symptoms, and its is often found incidentally in most reported cases (Davis et al. 1987; Yamamoto et al. 1979).

Most of the reported submucosal lymphatic cysts occurred in the small intestine, and submucosal lymphatic cysts in the esophagus (Brady and Milligan 1973), stomach (Fleming and Carlson 1970; Ramsay 1956; Yamamoto et al. 1979), and duodenum (Aase and Gunderson 1983; Aneiros et al. 1986; Davis et al. 1987; Matsui et al. 1985) were reported much less frequently.

In fact, there has been no reported case of a submucosal lymphatic cyst of the stomach in Korea. This fact prompted us to present a case of submucosal lymphatic cyst of the stomach which was confirmed by surgery.

CASE REPORT

A 46-year-old male visited Severance Hospital, Yonsei University College of Medicine because of abnormal upper gastrointestinal series findings which were taken two months previously at another hospital. The abnormal findings revealed a sharply circumscribed filling defect measuring 3×3 cm in size with normal overlying mucosa in the lesser curvature of the stomach (Fig. 1). The patient denied any symptoms or signs related with gastrointestinal disease.

The patient had been on an annual physical check-up program for the past several years. He worked in a trading company. He was a non-smoker and consumed a moderate amount of alcohol. He had no history of diabetes mellitus, allergy, hypertension, car-
diovascular disease, or tuberculosis.

On examination the patient appeared well. The body temperature was 36.4°C, pulse rate 80/min, respiration rate 20/min and blood pressure 120/80 mmHg. No rash or lymphadenopathy was found. The head and neck were normal, the lungs were clear, and the heart was normal. Abdominal examination was negative. The extremities were normal; no peripheral edema, clubbing, or cyanosis was found. The genitalia and rectum were normal.

CBC showed a hemoglobin of 14.7 gram per 100ml and a hematocrit of 40.2 percent. The white-cell count was 6500, with 53 percent neutrophils, 41 percent lymphocytes, and 6 percent monocytes. The platelet count was 196,000. The prothrombin time was 12.1 seconds, with a control of 12.1 seconds. The guaiac test was negative for occult blood and the urinalysis was within normal limits. Blood chemistries were as follows: urea nitrogen 21mg per 100ml, creatinine 1.2mg per 100ml, glucose 85mg per 100ml, uric acid 7.8mg per 100ml, bilirubin 1.2mg per 100ml, calcium 9.3mg per 100ml, phosphorus 3.7mg per 100ml, cholesterol 190mg per 100ml, and protein 7.6g (albumin 4.6g, globulin 3.0g) per 100ml.

The serum sodium was 140mmol, potassium 4.1mmol, chloride 104 mmol, and carbon dioxide 24mmol per liter. The serum aspartate aminotransferase (ASAT) was 25IU (normal, 8 to 30), serum alanine aminotransferase (ALAT) 33IU (normal, 8 to 30), lactate dehydrogenase (LDH) 64IU (normal, 52 to 127) and alkaline phosphatase 60IU per liter (normal, 30 to 115). The electrocardiogram and chest X-ray films were normal.

The fiberoptic panendoscopic examinations, done two times at the outpatient clinic and during the hospitalization, disclosed a cyst-like bulging mass, 3×3cm in size, in the lesser curvature of the gastric antrum (Fig. 2). The lesion was so soft that it was easily compressible with biopsy forceps. The biopsy was done two times with negative findings for malignant cells.

The results of a computed tomographic (CT) scan of the abdomen and a barium enema were within normal limits.

On the sixth hospital day, the abdomen was explored. Through a gastrotomy opening, an encapsulated cystic mass was excised from the submucosal layer. The cyst contained a dark brownish clear fluid. The liver, spleen, pancreas, and other intraperitoneal organs were grossly normal.

Microscopically, the endoscopic biopsy revealed a chronic nonspecific inflammation of the mucosa and did not include the submucosal layer. Microscopic examination of the excised cyst of the stomach disclosed dilated cysts at the submucosa. The cyst wall was composed simply of endothelial cells and a thin collagenous tissue. No muscle coat of its own was found. There were no red blood cells in the space of the cyst (Fig. 3).
DISCUSSION

A submucosal lymphatic cyst or lymphangioma is a thin-walled cyst which is lined by flattened endothelium. It has no septa but the surrounding layers of collagenous fibrous tissue often form a thin and incomplete capsule. The lesion is limited to the submucosal layer, and it does not affect the lymphatic system of the overlying mucosal layer. The content of the cyst is usually an amorphous homogenous or granular eosinophilic material (Aneiros et al. 1986; Shilkin et al. 1968).

In our case, the submucosal cyst of the stomach appeared to be dilated lymphatic spaces lined by endothelial cells. The lymphatic spaces had a thin collagenous wall without any muscle of their own. There were no red blood cells in the space of the cyst (Fig. 3).

The pathogenesis of this lesion is unknown, and there is no correlation with obstruction of the lymphatic system, unlike diffuse intestinal lymphangiectasia (Shilkin et al. 1968). Aase and Gunderson (1983) suggested that the pathogenesis might be correlated with the aging process of the intestinal wall because there was not a single case reported under 55 years of age among 35 cases of submucosal lymphatic cyst of the gastrointestinal tract. The age incidence of the 27 cases of lymphangioma of the stomach, reviewed by Yamaoto et al. (1979), was 51 years of age on the average.

The incidence of the submucosal lymphatic cyst is very low and it is uncommon to find it at sites other than the small intestine (Fujimiki et al. 1984; Syundo et al. 1986; Tanaka 1981; Waldman et al. 1961). A few cases were reported in the esophagus (Brady and Milligan 1973), stomach (Fleming and Carlson 1970; Ramsay 1956; Yamaoto et al. 1979) and duodenum (Aase and Gunderson 1983; Anerios et al. 1986; Davis et al. 1987; Matsui et al. 1985). Yamaoto et al. (1979) reviewed a case of lymphangioma of the stomach and also, reviewed 27 cases of the disease on the base of the literatures as reported during a 24-year period in Japan. Yao et al. (1981) reported only 3 cases of submucosal lymphatic cysts in their retrospective study of primary benign neoplasms of the jejunum and ileum during a ten-year period in Japan. Fleming and Carlson (1970) reported that of seven patients with single submucosal cysts, three of the cysts were located in the duodenum, two in the stomach, one in the jejunum, and one in the descending colon.

A submucosal lymphatic cyst rarely elicits systemic findings such as generalized edema, and hypoproteinemia; however, vague symptoms such as nausea, indigestion, and weight loss occurred in many cases (Davis et al. 1987; Dobbins 1983; Yamamoto et al. 1979). In 27 cases of lymphangioma of the stomach, reviewed by Yamamoto et al. (1979), the incidences
of the chief complaints were discomfort of the upper abdomen (37%), epigastric pain (18.5%), and no complaints (14.8%), in that order. In one case of submucosal lymphatic cyst of the stomach, reported by Fleming and Carlson (1970), vague dyspepsia was the only symptom. Laparotomy was undertaken to rule out a possible malignancy and finally a submucosal lymphatic cyst was confirmed. Ramsay (1956) reported a case which produced a clinical picture of hypertrophic pyloric stenosis due to a submucosal lymphatic cyst that occurred near the pylorus in the greater curvature in a baby aged 8 days old.

In our case, the patient did not experience any overt symptoms of gastrointestinal disease; moreover, the laboratory data did not give any remarkable clues.

On fiberoptic panendoscopic examination, the lymphatic cyst presents as a submucosal mass lesion with normal overlying mucosa. Likewise, leiomyoma, aberrant pancreas, intramural hematoma, hemangio- ma, isolated varix, dermoid cyst, or lipoma may have the same endoscopic appearance. The important point differentiating a submucosal lymphatic cyst from other lesions is that it is easily compressible by biopsych forceps and has a translucent cystic wall (Davis et al. 1987; Fleming and Carlson 1970). In our case, the endoscopic examination disclosed a 3×3cm sized cyst-like bulging mass, which was easily compressed with biopsych forceps, in the lower body of the antrum along the lesser curvature of the stomach (Fig. 2).

The endoscopic biopsy could be the most reliable and safest diagnostic modality for the submucosal lymphatic cyst. Brady and Milligan (1973) reported that a submucosal lymphatic cyst of the esophagus could be distinguished from a leiomyoma or a hemangioma of the esophagus by endoscopic visualization; thereafter, a biopsy could be performed without complications such as hemorrhage or infection.

A submucosal lymphatic cyst on the upper gastrointestinal series reveals a sharply defined, smooth, curvilinear filling defect with intact mucosa which often changes its shape readily in response to compression and peristaltic activity by fluoroscopy. However, this characteristic is also applicable to lipomas because of their fluid consistency at body temperature, but a lymphatic cyst can be differentiated from a lipoma which is bounded by a fuzzy ill-defined margin and has an ulcerated overlying mucosa (Fleming and Carlson 1970). Judging from the findings of the gastrointestinal series and/or fiberoptic panendoscopy, it seems safe to assume that (1) changes of the shape of gastric submucosal tumors should not only be more carefully pursued roentgenologically, but also (2) confirmed endoscopically, and (3) the presence of fluctuation of the tumor should also be detected endoscopically for the exact preoperative diagnosis of cyst of the stomach (Yamamoto et al. 1979). In our case, the findings of the upper gastrointestinal series revealed a 3×3cm sized, sharply circumscribed filling defect with normal overlying mucosa on the lesser curvature of the stomach. But we did not certify the change of shape in response to compression and peristaltic activity because we did not repeat the upper gastrointestinal series.

In treatment of the submucosal lymphatic cyst of the stomach, the main feature of practical importance is the intimate relation of the cyst to its surrounding structures morphologically. The cyst shares a common wall with the stomach and the two cannot easily be dissected apart. This fact has an important bearing on treatment because it makes simple enucleation impossible and accounts for the variety of methods that have been tried. These include: (1) aspiration; (2) local excision; (3) marsupialization; (4) excision of the septum between the cyst and the lumen of the stomach; and (5) gastro-enterostomy (Ramsay 1956). In our case, treatment of the submucosal lymphatic cyst of stomach was done by local excision and the cyst could easily be dissected apart.

CONCLUSION

A submucosal lymphatic cyst of the stomach is extremely rare. Its pathogenesis is unknown and it is incidentally found in most cases. We would like to report a case of submucosal lymphatic cyst of the stomach which was confirmed after exploration.

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