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Case Report

Foregut duplication cyst of the stomach

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Foregut duplication cyst of the stomach is an extremely rare disease entity. A 35-year-old Korean man presented with epigastric pain. An abdominal cystic mass, measuring 7 × 6 × 5 cm, was found in the lesser curvature of the stomach. The cyst was unilocular with a grey-white, rubbery wall. Microscopically, the cyst wall was lined by pseudostratified ciliated, columnar epithelium and gastric mucosa with a complete lining of smooth muscle bundles. Although the origin of this lesion remains uncertain, this case suggests that the gastric cyst arose from the embryonic foregut and showed differentiation toward respiratory and gastric structures.

Key words: foregut duplication cyst, stomach

Cystic lesions of the stomach are uncommon and generally comprise heterogeneous groups such as developmental anomalies and post-traumatic, infectious, and neoplastic lesions. A gastric duplication cyst is considered to be a cystic form of gastric duplication, which stems from a congenital anomaly. Gastric duplication cysts are uncommon lesions that represent only 2–8% of all alimentary duplication cysts.[1,2] Although several cases of a gastric cyst lined by pseudostratified ciliated columnar epithelium (PCCE) have been reported,[3–6] only two cases of gastric duplication cysts containing both gastric mucosa and PCCE, located in the greater curvature of stomach, are reported in the English literature.[5,6]

A unique example of non-communicating, foregut duplication cyst (FDC) in the lesser curvature of the stomach, which is lined by gastric and respiratory type mucosa is reported.

CLINICAL SUMMARY

A 35-year-old man presented with a 10-year history of epigastric discomfort with worsening epigastric pain for the last 4 months. There was no history of nausea, vomiting, fever, jaundice, change in bowel habit, abdominal surgery, or other significant medical conditions. Physically, he was a well-nourished, healthy looking young man. He complained of moderate epigastric tenderness without rebound tenderness, shifting dullness, or definitive palpable mass. Laboratory evaluation was unremarkable with respect to blood count, serum amylase level, liver function test and urinalysis.

Chest and abdominal X-rays were unremarkable. An abdominal sonography and computed tomography scan revealed a 7 cm cystic lesion between the lesser curvature of the stomach and the left lobe of the liver (Fig. 1). It showed a homogeneous low density with a thin wall. Neither solid part nor mural nodules was seen. There was a 1 cm-sized hemangioma in the right lobe of the liver. Initial radiological diagnosis showed either a duplication cyst of the stomach or a simple cyst of the liver. The patient underwent fiberoptic gastroscopy. The wall of the stomach revealed an externally compressed mass lesion on the lesser curvature side of high body.

At surgery, a large cystic lesion in the lesser curvature aspect of the stomach from the antrum to the cardio-esophageal junction was found. The cystic lesion had no connection with the liver, but showed a partial adhesion to the stomach. There was no communication between the duplication cyst and the lumen of stomach. Cystectomy was performed without any difficulty and the patient had an uneventful recovery.

PATHOLOGICAL FINDINGS

The specimen obtained was a 7 × 6 × 5 cm-sized unilocular cystic mass (Fig. 2). The rubbery walls were 0.4–0.8 cm in thickness with no solid portion. The cyst content measured approximately 80 mL of thick brown fluid with a small amount of sludge-like material. The inner lining was smooth, gray-white in color with partial erosion or ulceration.

For light microscopic examination, the specimen was fixed with 10% buffered formalin, embedded in paraffin and

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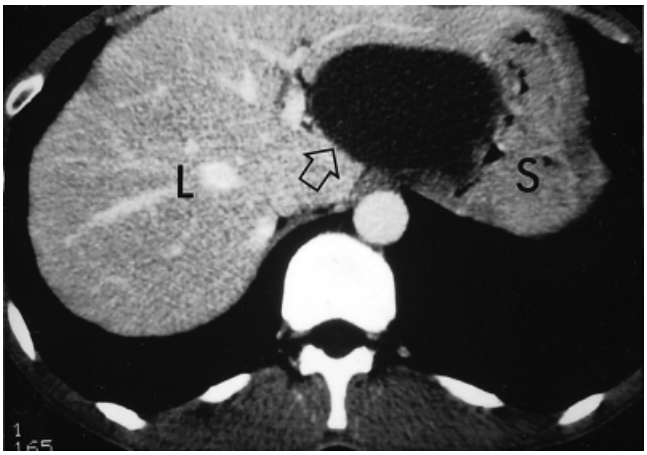

Figure 1 Computed tomography of the abdomen showing a well-circumscribed cystic mass (open arrow). The cyst is located between the lesser curvature of stomach (S) and the liver (L).

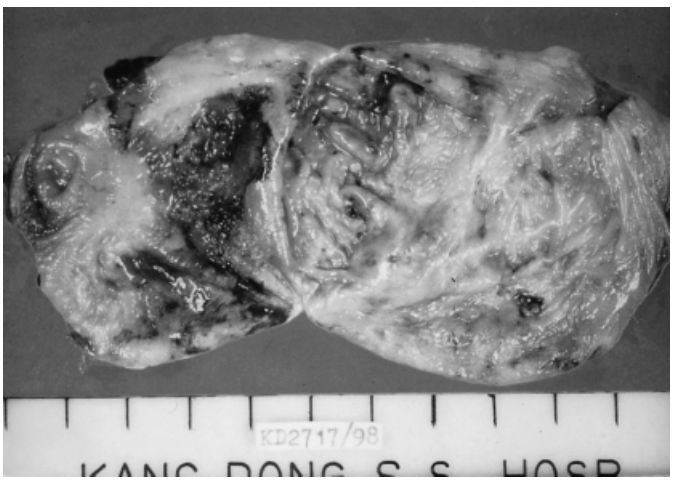

Figure 2 The inner surface of the cystic mass is smooth, gray-white in color with partial ulceration.

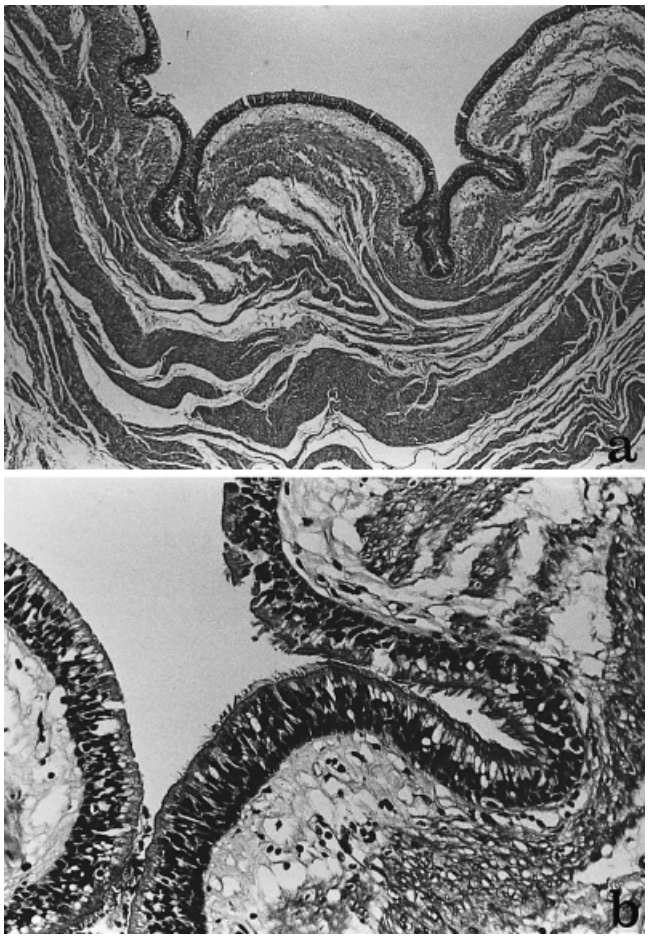

Figure 3 (**a**) Photomicrography of the cyst wall shows pseudostratified ciliated columnar epithelium and smooth muscle. (**b**) High power view demonstrates fine cilia on the luminal surface of lining cells (HE).

processed routinely. Microscopically, the cyst wall consisted of mucosa, subepithelial connective tissue, and two to three layers of smooth muscle with an outermost thin fibrous capsule. The largest portion of the mucosa was lined by PCCE, which comprised approximately 60% of the cyst (Fig. 3). About 20% of the lining epithelium consisted of gastric foveolar epithelium with fundic or pyloric glands (Fig. 4). Other areas showed erosion or ulceration accompanied by inflammatory cell infiltration.

DISCUSSION

Gastrointestinal duplications can occur anywhere throughout the gastrointestinal tract, from the mouth to the anus.[7,8] They are most commonly found in the ileum, whereas stomach is rare. In a review of 109 gastric duplications,[1] the majority of the patients were in the first 3 months of life, and 63.3% were female. Gastric duplications vary in size and shape, although most were 3–6 cm in size. Most cases showed tubular or cystic structures with mucosal linings that ranged from a simple columnar epithelium to normal-appearing gastric or intestinal mucosa. A malignant change within a gastric duplication cyst was rarely found.[2]

Our case showed a duplication cyst containing both PCCE and gastric mucosa. Four cases of gastric cysts with respiratory lining epithelium have, to our knowledge, been reported,[3–6] and the main features of the cysts are summarized in Table 1. The two cases showed intramural cysts that had a PCCE lining epithelium without gastric mucosa.[3,4] Two reports of gastric duplication cyst containing both gastric and respiratory mucosa have been demonstrated,[5,6] both were located in the greater curvature of the stomach. The majority of the duplications are distributed dorsal to the primitive gut developmentally.[9] Thus, a gastric duplication cyst usually

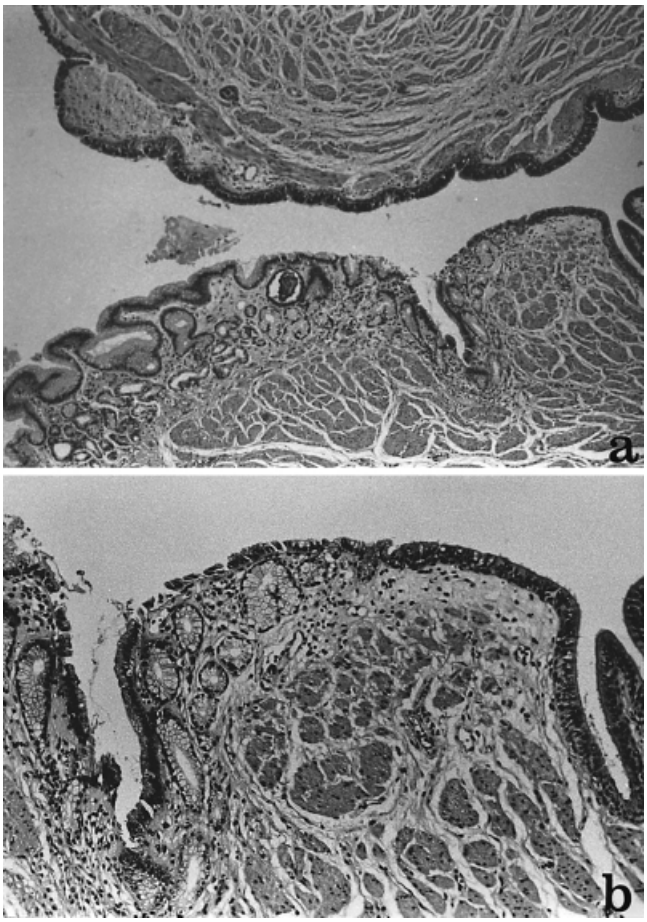

Figure 4 (**a**) Transitional area of pseudostratified ciliated columnar epithelium to gastric mucosa. (**b**) High-power microphotograph of transitional area (HE).

appears along the greater curvature of the stomach, adjacent to the gastric wall. In the embryonic stage, persistence of a vacuole formed in solid phase, which lies chiefly near the lesser curvature of the stomach, may be an origin of duplication.[10] However, Bremer[11] postulated that the stomach does not go through a solid stage. Although the exact mechanism of gastric duplications other than those of greater curvature is uncertain, 5.5% of gastric duplication cysts were actually located in the lesser curvature of the stomach.[1] The present case is unusual as the cyst was located in the lesser curvature of the stomach and contained both gastric and respiratory epithelium.

The mucosal lining of duplication may be histologically similar to that of the segment of the gut to which it is topographically related. However, some mucosa of duplications may intimate any other segment of alimentary or respiratory tract.[12] Duplication cysts lined by ciliated columnar epithelium were also found in the alimentary tract, which was located in the medial wall of the ascending colon.[13] Mathur *et al.*[14] reviewed eight cases of thoracic and five cases of abdominal duplications in which a thoracic cyst was lined by PCCE and gastric mucosa. Ohbayashi *et al.*[8] reported a duplication cyst of the tongue lined by stratified squamous and respiratory type epithelium. The PCCE was also found in a cyst of the liver which was derived from the primitive foregut.[15] The presence of respiratory epithelium in the cysts of the thorax, tongue, liver, and stomach suggested that the undifferentiated epithelium of the foregut might undergo transition to differentiated, specialized epithelium during the embryonic period.

Table 1 Summary of gastric duplication cyst lined by respiratory mucosa

No	Sex	Age (years)	Location	Size (cm)	Lining cell	Subepithelial content	Ref.
1	F	46	Intramural, Greater curvature, Fundus	8 × 6	PCCE Squamous metaplasia	Subepithelial connective tissue Smooth muscle	3
2	F	61	Intramural, Cardia	6	PCCE Ulcer (+)	Subepithelial connective tissue Mature cartilage Seromucinous gland Smooth muscle	4
3	F	35	Greater curvature	5.5 × 2.5 × 2	PCCE Gastric epithelium	Submucosa Smooth muscle	5
4	M	25	Subserosal, Greater curvature, Fornix	6.5 × 5 × 5	PCCE, ulcer (+) Gastric epithelium Squamous and intestinal metaplasia	Smooth muscle	6
5	M	35	Lesser curvature	7 × 6 × 5	PCCE Gastric epithelium Ulcer (+)	Subepithelial connective tissue Smooth muscle Fibrous capsule	Present case

PCCE, pseudostratified ciliated columnar epithelium.

Duplications result from disturbances in embryonic development and various theories have been proposed as to the actual mechanism. Bremer[11] proposed the theory of errors of recanalization and fusion of longitudinal folds. He suggested that duplication cysts originated from the fusion of longitudinal folds, allowing the passage of a bridge of submucosa and muscle at the second and third months of intra-uterine life. McLetchie[16] suggested that adhesion of notochord and embryonic endoderm might not elongate as quickly as its surrounding structures, causing traction diverticulum leading to duplication cyst formation. Other theories of enteric duplication include abortive twinning,[17] persistent embryological diverticula,[18] and hypoxic or traumatic events that cause a duplication.[19] But, there is no single theory that is satisfactory for all types of duplication.

Embryologically, the primitive foregut gives rise to the pharynx, esophagus, respiratory tract, stomach, proximal part of duodenum, and the hepatobiliary system, and it is possible that these areas may be subject to duplication located in the stomach. During the early embryonal stage of foregut development, malformations such as duplications may result from a supernumerary lung bud found in the foregut during the fifth and seventh week of embryogenesis.[20] Gensler *et al.*[3] suggested that the duplication cyst with PCCE was derived from the caudad-most portion of the laryngotracheal outgrowth, which remained attached to the portion of the primitive foregut destined to become the stomach. Arrests at various stages of the separation of the laryngotracheal groove and the premature foregut may result in different congenital anomaly, such as tracheoesophageal fistula. Therefore, it suggests that the FDC of the stomach may arise as a detached outpouching of the primitive foregut.

Associated anomalies are found in about 50% of gastric duplication cysts.[1] The most common anomaly is duplication of the esophagus, followed by vertebral anomalies. Although it is unclear whether the hemangioma is an associated anomaly or an incidental combined lesion, the hemangioma, such as in this case, is rarely combined. The present case revealed a partial erosion or ulceration on the mucosal surface. This might be due to digestive enzyme secreted from gastric glands that have destroyed a bronchial epithelium and resulted in ulceration,[6] or to ischemic change by vessel distortion.[21]

In summary, a case of gastric duplication cyst in a 35-year-old man is presented. This is the third case of a gastric foregut duplication cyst containing both gastric and respiratory mucosa, and is located in an unusual site, the lesser curvature of the stomach.

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