

Bronchogenic Gastric Cyst. A Case Report

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Abstract. A case of congenital bronchogenic cyst in the gastric mucosa is presented. The cyst was lined by pseudostratified epithelium and covered with ciliated cells. Congenital bronchogenic cysts should be differentiated from acquired gastric cysts lined with ciliated metaplastic cells that evolve as a result of environmental factors.

Bronchogenic cysts are congenital anomalies evolving from the ventral foregut between the 3rd and the 7th prenatal weeks. Bronchogenic cysts are lined with cuboidal or pseudostratified ciliated epithelium and may or may not be surrounded by elastic fibers, smooth muscle and cartilage.

Bronchogenic cysts are divided into intra-thoracic or supra-diaphragmatic and intra-abdominal or sub-diaphragmatic (1). Sub-diaphragmatic bronchogenic cysts are rare, particularly those located within the confines of the gastric wall exclusively. Despite the fact that Gensler *et al.* (1) described the first case nearly 50 years ago, only two additional cases have been reported (2,3).

Recently, we found a new case of bronchogenic cyst in the gastric mucosa. The purpose of this report was not merely to describe an additional case, but to draw attention, for the first time, to an important differential diagnosis between intramucosal congenital cyst and acquired intramucosal cysts also lined with ciliated cells (4).

Case Report

A 26-year-old Swedish male consulted because of periodic epigastric pain. The pain had begun 18 months previously and he had been periodically medicated with proton pump inhibitors. Palpation evoked epigastric

pain. It was assumed that the symptoms were induced by gastroesophageal reflux. Manometry revealed normal peristalsis in the entire esophagus, with normal tonus as well as normal relaxation of the lower esophageal sphincter. The pH was also normal. A gastroscopy showed signs of mild distal esophagitis; no gastric inflammation or hiatus hernia were present. The pinch biopsies taken revealed grade 1 esophagitis and normal gastric mucosa without *H. pylori*. One of the biopsies from the corpus, however, showed an intramucosal cyst. A closer examination revealed that the intramucosal cyst was lined with pseudostratified epithelium built with cuboidal cells (Figure 1), some of them being vacuolated. In the immediate *lamina propria mucosa* underneath, a lymphatic follicle was found. No cartilaginous tissue was present. No signs of inflammation, intestinal metaplasia or epithelial dysplasia were found in the adjacent fundic mucosa.

The cells lining the luminal aspect of the cyst had, on their free border, densely packed cilia of the same height (Figure 1). Each cilium was approximately 6 µm long. Sections were stained with PAS, PAS-diastase, Tubulin B (Sigma-Aldrich, Sweden) and Ki67 (Clone MIB1, Dako, Glostrup, Denmark). Only one PAS diastase-resistant clear cell was found. The aforementioned vacuolated cells were PAS-negative. The cilia were positive for Tubulin B immunostain (Figure 2) and showed no signs of cell proliferation; only one intraepithelial lymphocyte was MIB1-positive.

The patient received omeprazol medication and the symptoms disappeared.

Discussion

Only three cases of congenital bronchogenic cysts confined to the boundaries of the gastric mucosa have been reported (1-3).

Other sub-diaphragmatic bronchogenic cysts, not restricted to the gastric wall but connected with the gastric mucosa, are also on record. Braffman *et al.* (5) found one case of broncho-pulmonary foregut malformation in the left subphrenic space that communicated with the gastric

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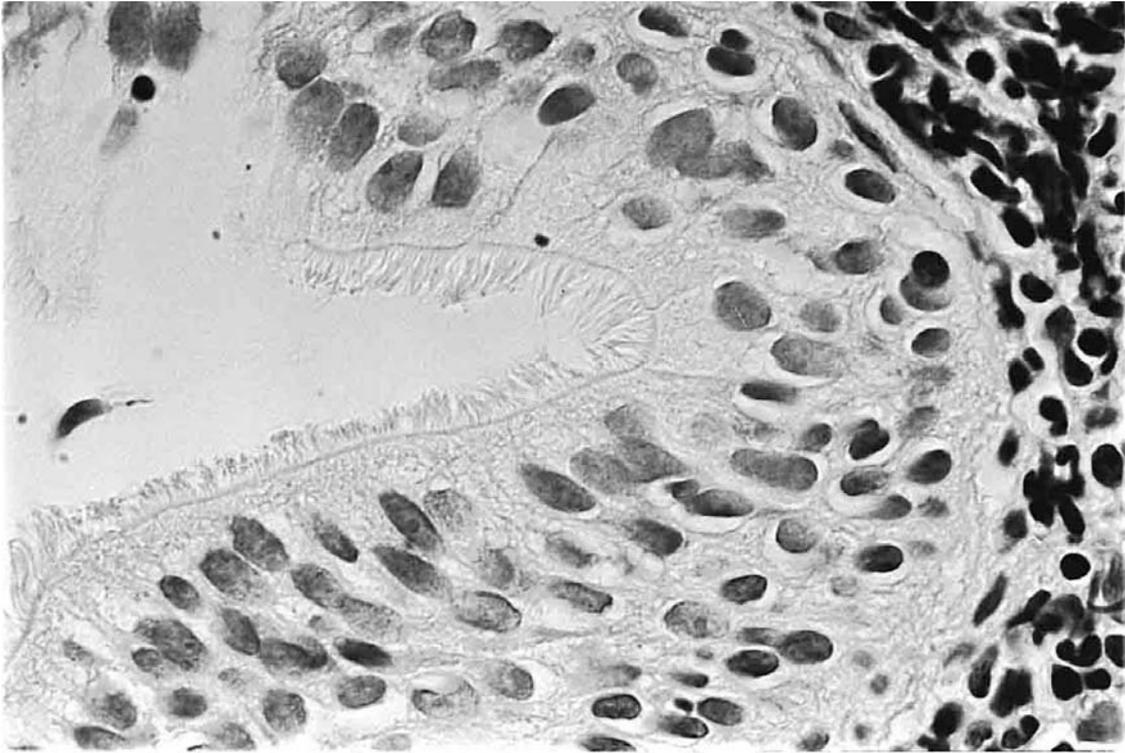


Figure 1. Detail from a bronchogenic gastric cyst, showing ciliated pseudostratified epithelium (H&E, 50x).



Figure 2. Bronchogenic gastric cyst showing cilia stained with Tubulin B (25x).

fundus. Keohane *et al.* (6) reported a subdiaphragmatic bronchogenic cyst with communication to the stomach and Takashita *et al.* (7) also found a communication with the gastric fundus in one of two cases of abdominal bronchogenic cysts. More recently, Matsubayashi *et al.* (8) found one case of subphrenic bronchopulmonary foregut malformation having mature lung tissue with randomly distributed bronchial structures and ciliated epithelium-lined cysts, some of which were lined with gastric mucosa.

The histological lining of the bronchogenic cyst here reported is at variance with that of other subtypes of intramucosal gastric cysts, also lacking cartilage—namely of foveolar, fundic, pyloric, intestinal metaplastic and ciliated metaplastic phenotypes (4). Gastric mucosal cysts with ciliated metaplasia are lined with a single row of gastric seromucinous cuboidal cells or with intestinal metaplastic goblet cells having on their free border irregular, sparse cilia. Those acquired ciliated cysts evolve as a result of environmental factors (9), and are usually present in specimens from Asian patients having elsewhere a gastric carcinoma of intestinal histological type (10). Metaplastic ciliated cysts are usually located in the antrum, but they may also be found in the cardia and in the corpus in cases with extended intestinal metaplasia (9).

Bronchogenic cysts are very rare. A similar cystic phenotype was not detected in two large series comprising gastric biopsies from 1675 Japanese, Mexicans and Swedish patients (11) or resected stomachs from 3406 patients dwelling in disparate geographical regions of the Atlantic and the Pacific Basins (9).

In conclusion, a case of congenital bronchogenic cyst in the gastric mucosa has been reported. Bronchogenic cysts should be differentiated from acquired gastric ciliated cysts evolving as a consequence of environmental factors (9).

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