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Case report

Bronchogenic cyst in the abdomen

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Summary. A bronchogenic cyst was found in the abdomen, in the retroperitoneum adjacent to the superior pancreatic body. The cyst was unilocular and contained about 100 ml pale yellow mucinous fluid. Microscopic examination revealed a pseudostratified columnar ciliated or cuboidal epithelium, seromucous glands, smooth muscle and cartilage, the distinctive features of bronchogenic cysts. This aberrant location of the cyst is explicable if abnormal buds of the tracheobronchial tree are pinched off and migrate into the abdomen in an early embryonic stage before the canal linking the abdominal with the thoracic cavity is closed by fusion of the future components of the diaphragm.

Key words: Bronchogenic cyst – Abdomen – Retroperitoneum

Introduction

Bronchogenic cysts are rare congenital anomalies usually occurring in the pulmonary parenchyma or in the mediastinum (Maier 1948; Rogers and Osmer 1964). They are rarely found in aberrant locations such as the skin and subcutaneous tissue in the vicinity of the sternal region, shoulder and neck (Seybold and Clagett 1945; Fraga et al. 1971), intrapericardium (Dabbs et al. 1957; Deenadayalu et al. 1974), intra-diaphragmatic (Aaron 1965; Brickman et al. 1982) and intradural cervical (Yamashita and Harris 1973). However, such a cyst located in the abdomen is extremely rare and only two cases have been described in the literature (Miller et al. 1953; Murley and Lenz 1979).

We now report a bronchogenic cyst occurring in the abdomen, the retroperitoneum adjacent to the pancreas. The embryogenesis of this anomaly is discussed.

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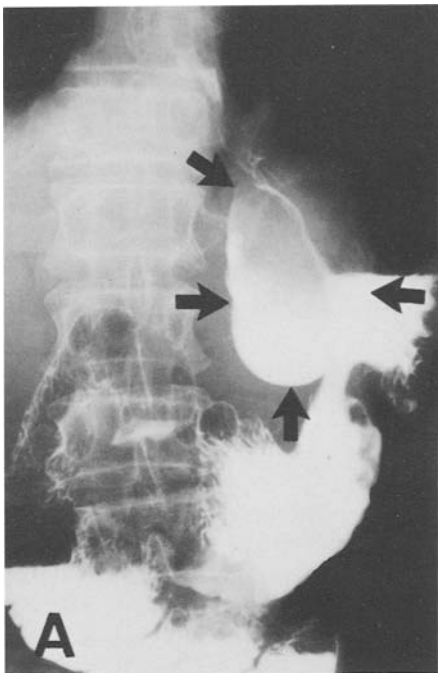

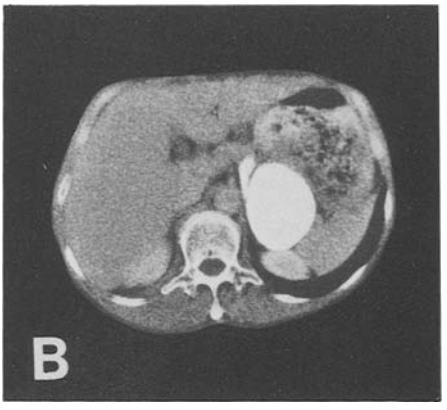

Fig. 1. A Percutaneous cystography and upper GI series. An egg-shaped cyst (*arrow*) is evident behind the body of the stomach. **B** Percutaneous cystography and CT scan. The cyst with a short duct is located in the retroperitoneum adjacent to the stomach and to the spleen

Fig. 2. Macroscopic features of the opened cyst. The inner smooth surface is roughly trabeculated

Case report

The patient, a 59-year-old Japanese man, was admitted because of epigastric pain accompanied by nausea and vomiting. He had recurring episodes of these symptoms during the past four years.

CT scan and echogram revealed a cystic mass, about 6 cm in diameter, situated in the retroperitoneum adjacent to the dorsal aspect of the body of the stomach and medial to the spleen. Endoscopic retrograde pancreaticography showed no continuity of the pancreatic

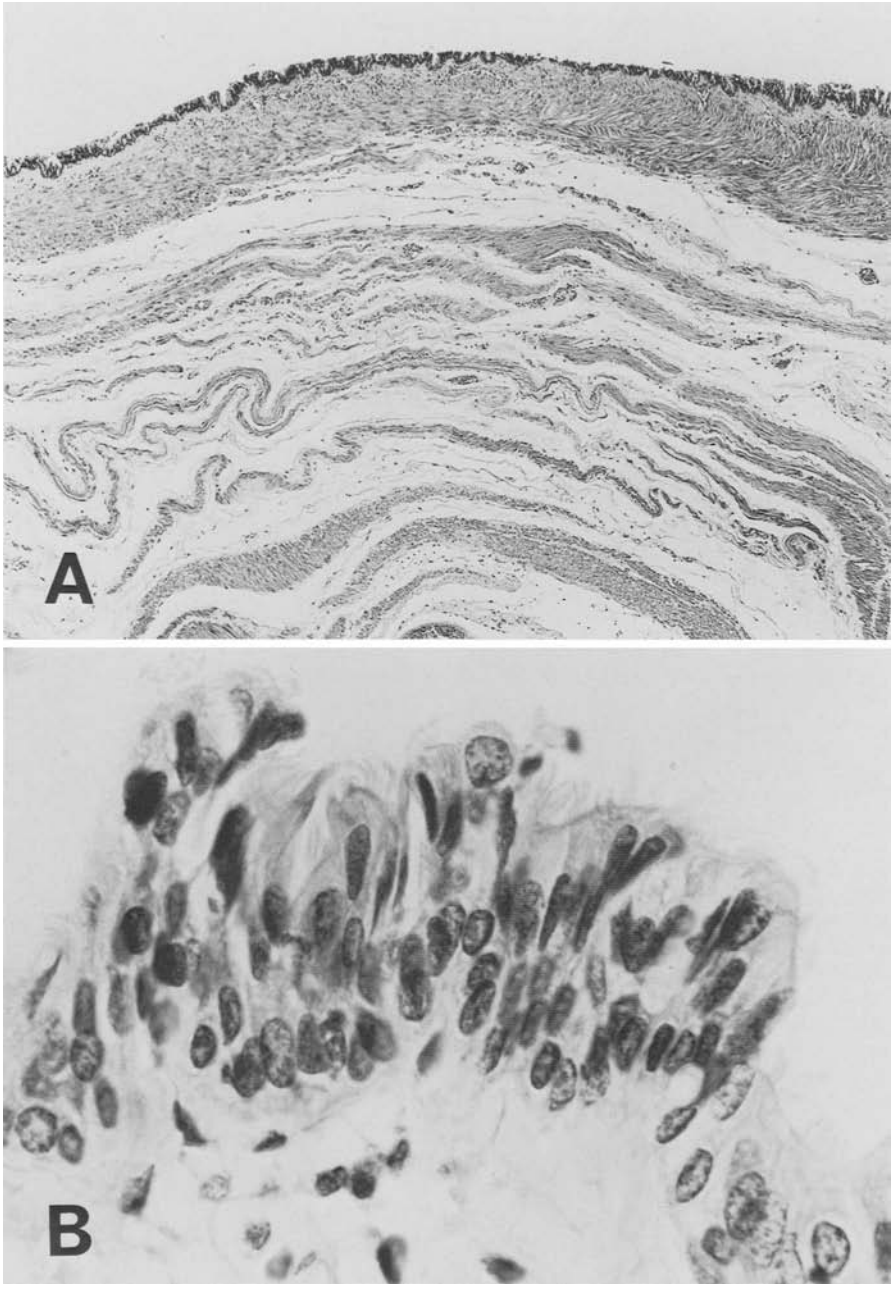

Fig. 3. A The wall of a cyst lined with pseudostratified columnar ciliated epithelium and smooth muscle fibers. (H & E ×48). **B** High power view of pseudostratified columnar ciliated cells lining the cyst. (H & E ×920)

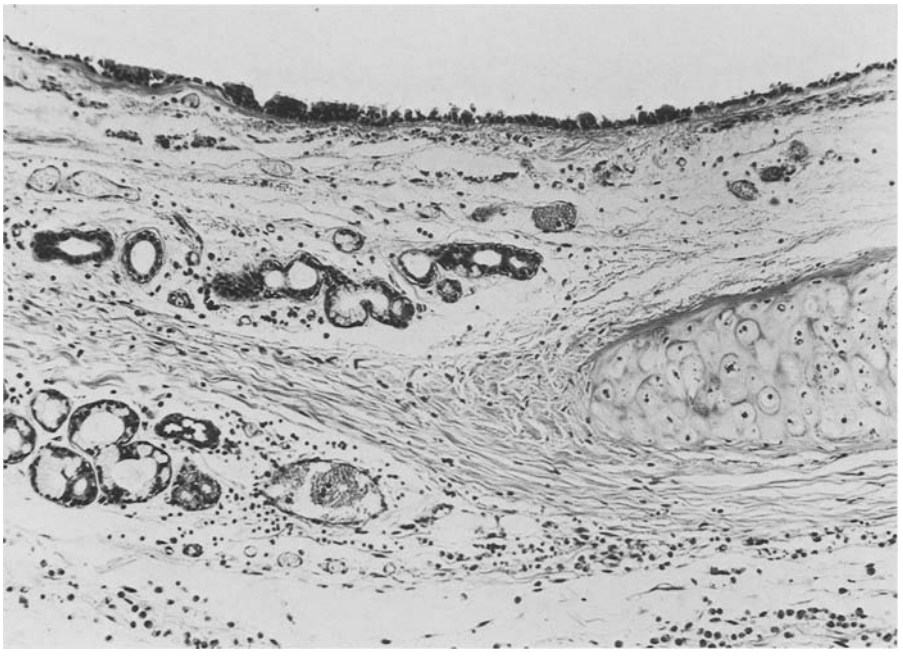

Fig. 4. Cartilage and seromucous glands in the cyst wall. (H & E ×114)

duct with the cavity of the cyst. Percutaneous cystography confirmed an egg-shaped cyst and disclosed no communication of its cavity with other structures. At the same time, CT scan and upper GI series were carried out to identify the contour and location of the cyst more clearly (Fig. 1A and B). Aspiration of the cyst contents revealed elevated levels of amylase and elastase 1, 13,700 U/dl and 10,000 ng/ml, respectively.

Laparotomy revealed a fluctuating cyst with a smooth outer surface and measuring about 7 cm in diameter. It was pale blue and located in the retroperitoneum adjacent to the superior pancreatic body and medial to the spleen, displacing the left gastric artery to the right. It was attached to the pancreas by loose fibrous connective tissue. A short winding duct extended from the superior pole of the cyst toward the diaphragm and ended before reaching it. However, there was no duct or fistula connecting the cyst with the pancreas or other structures. Extirpation of the cyst was performed without difficulty, and in addition, a band of fibrous tissue connecting the transverse colon and ileum, probably causing ileus, was excised. Except for the cyst, no other anomalies were evident. The postoperative clinical course was satisfactory.

Pathologic findings

Grossly the cyst was unilocular and contained about 100 ml of pale yellow, mucinous fluid. When opened, the thin cyst wall measured 5.5 × 6.5 × 0.5 cm. The inner surface was generally smooth but roughly trabeculated and was yellowish brown (Fig. 2). There was adipose tissue only on the smooth outer surface.

The wall was wholly lined by a layer of pseudostratified columnar ciliated and partly cuboidal cells with a lamina propria underneath (Fig. 3A and B). Exterior to the latter was loose fibrous connective tissue and sparse

bundles of smooth muscle fibers intermingled with small vessels and a small number of nerves. Elastic fibers were relatively abundant within and around the bundles of smooth muscle fibers. Hyaline cartilage and seromucous glands, both identical to those in the bronchus, were present in part within the wall of the cyst (Fig. 4). A few bundles of skeletal muscle fibers surrounded the short winding duct. Pancreatic tissue, pulmonary alveoli or other tissues suggestive of a teratomatous origin were absent.

Discussion

Adams and Thornton (1943) described bronchogenic cysts of the mediastinum as follows: the cysts very thin-walled and are filled with a thin, milky-colored mucoid material. Histologically, the smooth lining consists of flattened to columnar ciliated epithelium and the wall contains fibrous tissue, smooth muscle, mucous glands, cartilage, nerves, and blood vessels. In the current case, identification of the lesion as a bronchogenic cyst was not difficult as the histological features were distinct even though the retroperitoneum is an extremely rare location for such a cyst. In addition, the macroscopic appearance was consistent with that of a bronchogenic cyst.

Bronchogenic cysts are formed as a result of abnormal budding and pinching off of the tracheobronchial tree at about the fifth week of intrauterine life, a time when bronchial buds develop to form the primitive respiratory tree. Although the derivatives are usually located in the vicinity of the bronchus and the trachea in the mediastinum, they may migrate to an aberrant location if connection with the tracheobronchial tree is lost (Fraga et al. 1971). However, the aberrant locations so far reported are restricted to the upper portion of the body, such as chest and neck.

A possible explanation for occurrence of the cyst in the abdomen is as follows: the thoracic and abdominal cavity are linked by the pericardioperitoneal canal in an early embryonic stage, and when the canal is divided into two respective cavities by fusion of the pleuroperitoneal membranes, that is, the future components of the diaphragm, at the end of the sixth week of intrauterine life (Moore 1982), abnormal buds of the tracheobronchial tree are pinched off by these membranes and migrate into the abdomen. In the current case, the presence of a short duct extending from the cyst toward the diaphragm was reminiscent of a previous connection of the cyst with the tracheobronchial tree.

We found only two other intra-abdominal bronchogenic cysts described in the literature. The first was among the nine cases of bronchogenic cyst reported by Miller et al. (1953). The occurrence was in an infant with multiple congenital anomalies including a duplicate stomach and a congenital heart lesion. Histologically, ciliated columnar epithelium was reported to line the cyst. The second case in a 17 year old boy was reported by Murley and Lenz (1979). The intra-abdominal cyst adhered to the diaphragm and oesophagus, with histological features including ciliated pseudostratified columnar lining cells and well differentiated hyaline cartilage as the components of the wall. Our findings differ in that smooth muscle fibers, seromucinous

glands and nerve fibers were evident, these being common components of a usual bronchogenic cyst, and that neither complicating anomalies nor adhesions to the diaphragm or oesophagus were recognized. As to a few bundles of skeletal muscle encountered in the current cyst, Frage et al. (1971) noted the same tissues in the cyst wall in three of their 30 patients with bronchogenic cysts in the skin and subcutaneous tissue.

Why the levels of amylase and elastase 1 in the cyst fluid were elevated was not elucidated. No ductal structure connecting the cavity of the cyst with the pancreatic duct was identified nor was there pancreatic tissue in the cyst wall. The presence of heterotopic pancreatic tissue in bronchogenic cyst has been reported (Jaschke et al. 1982).

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