

## RETROPERITONEAL PERIPANCREATIC BRONCHOGENIC CYST MIMICKING PANCREATIC CYSTIC TUMOR

Majda Vučić, Davor Tomas, Mario Zovak<sup>1</sup>, August Mijić<sup>1</sup> and Božo Krušlin

Ljudevit Jurak University Department of Pathology and <sup>1</sup>University Department of Surgery, Sestre milosrdnice University Hospital, Zagreb, Croatia

**SUMMARY** – Retroperitoneal cystic tumors are rare and among them retroperitoneal bronchogenic cysts are extremely infrequent. Bronchogenic cyst is a development abnormality of the primitive foregut, which typically occurs in the chest. We describe a rare case of isolated peripancreatic bronchogenic cyst in a 53-year-old man that presented with intermittent epigastric pain. Abdominal ultrasonography and computed tomography showed a cystic lesion closely related to the superior border of the pancreatic tail. The cystic tumor of pancreas was suspected. The cystic lesion was surgically removed and submitted for histopathologic analysis. Microscopically, the cyst was partially lined with pseudostratified, columnar, ciliated respiratory-type epithelium and partially with a single layer of tall columnar epithelial cells interspersed occasionally with goblet cells, which resided on the fibromuscular wall that contained seromucinous glands, and focally dense inflammatory cells. The diagnosis of bronchogenic cyst was established. Although rare in this region, bronchogenic cyst should be considered on differential diagnosis of peripancreatic cystic lesions.

**Key words:** *Bronchogenic cyst – complications; Bronchogenic cyst – diagnosis; Pancreatic neoplasms – pathology; Pancreatic neoplasms – diagnosis; Retroperitoneal neoplasms – diagnosis; Retroperitoneal neoplasms – pathology; Retroperitoneal neoplasms – surgery*

### Introduction

Bronchogenic cysts are congenital foregut abnormalities resulting from embryological budding of the bronchial tree. Generally, they are found in the posterior part of the mediastinum, particularly posterior to the carina and intrapulmonary regions<sup>1,2</sup>. However, they can rarely occur in such unusual sites as the skin, subcutaneous tissue, pericardium, and even more rarely in the retroperitoneum<sup>3</sup>. Subdiaphragmatic bronchogenic cysts usually are asymptomatic and are mostly an incidental finding. In the peripancreatic region, bronchogenic cysts are extremely rare and clinically and radiologically mimic pancreatic pseudocysts or cystic tumors<sup>4</sup>. The recommended treatment for bronchiogenic cyst is surgical

extirpation, to establish the diagnosis, remove the symptoms and prevent complications<sup>5</sup>. We report a case of a 53-year-old man with retroperitoneal bronchogenic cyst in the peripancreatic region and a history of intermittent epigastric pain.

### Case Report

A 53-year-old male patient was referred to our hospital with a one-year history of vague discomfort, intermittent pain and heat in the epigastric region. The patient was without previously history of illness. Physical examination demonstrated no palpable mass in the abdominal region. Results of laboratory tests, including blood counts and biochemical tests were within the normal limits. Abdominal ultrasonography showed a cystic mass adjacent to the pancreas. Computed tomography revealed a well-defined and circumscribed cystic mass with dense fluid content, which measured 8x6x6 cm (Fig. 1). Radiological findings did not show definitive demar-

Correspondence to: *Majda Vučić*, Ljudevit Jurak University Department of Pathology, Sestre milosrdnice University Hospital, Vinogradska c. 29, HR-10000 Zagreb, Croatia  
E-mail: [mvucic@kbsm.hr](mailto:mvucic@kbsm.hr)

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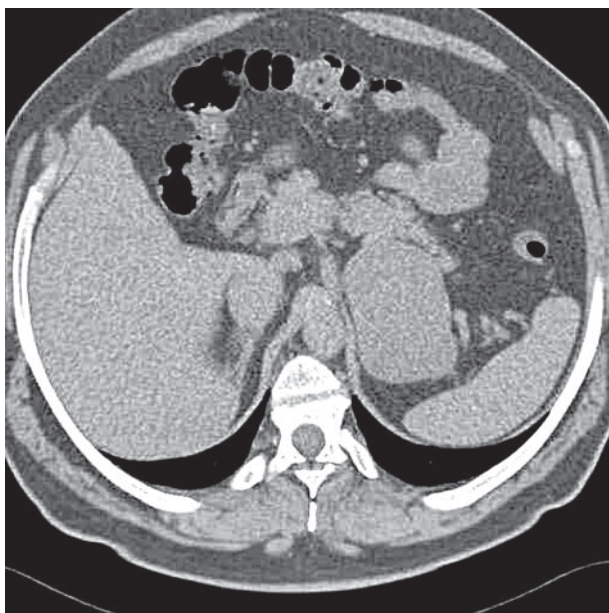


Fig. 1. Computed tomography revealed a well-defined and circumscribed cystic mass closely related to the superior border of the pancreatic tail.

relation between the mass and the pancreas, and the possible malignancy could not be ruled out. The patient underwent surgery for resection of the peripancreatic cystic mass. On laparotomy a cystic tumor was detected. It was related to the superior border of the pancreatic tail with extension cranially behind the stomach toward the hilus of the spleen. There was no attachment of cystic mass to the pancreas or any other struc-



Fig. 2. The cyst was unilocular and measured 7.5x4.5x0.7 cm. Outer surface was smooth and glistening. Inner surface was dark gray to brown, mostly smooth with focal thickening.

ture, allowing for simple enucleation. The entire cyst was removed and submitted for frozen section analysis. Grossly, the cyst was unilocular and measured 7.5x4.5x0.7 cm. Outer surface was smooth and glistening. Inner surface was dark gray to brown, mostly smooth with focal thickening (Fig. 2). Microscopic examination of the cyst on frozen section and paraffin embedded slides stained with hematoxylin-eosin revealed that the cyst was partially lined with pseudostratified, columnar, ciliated respiratory-type epithelium and partially with a single layer of tall columnar epithelial cells interspersed occasionally with goblet cells (Fig. 3A). The cyst wall

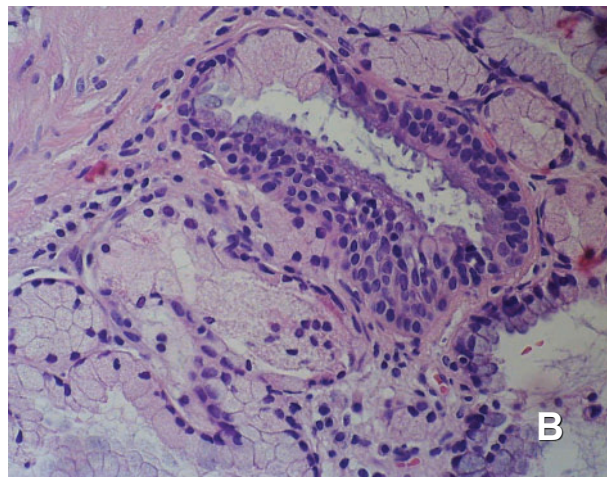
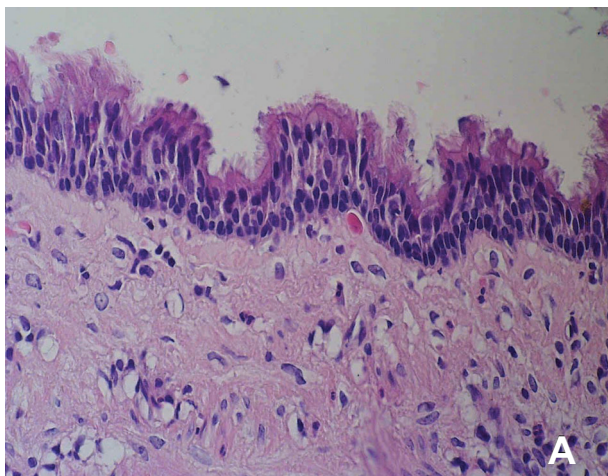


Fig. 3. The cyst was partially lined by pseudostratified, columnar, ciliated respiratory-type epithelium (A), and the wall was made of fibrovascular, connective tissue and a single layer of smooth muscle cells with dispersed seromucinous glands (B).

was made of fibrovascular, connective tissue and a single layer of smooth muscle cells with dispersed seromucinous glands and focally dense inflammatory infiltrate (Fig. 3B). The inflammatory cells were mainly lymphocytes, occasionally organized in follicles with germinal center, but in some areas histiocytes and foamy macrophages predominated. No sign of malignant transformation could be seen. The histologic picture, additional histochemical special stains (Alcian- Periodic acid-Schiff) and immunohistochemical analyses for S 100,  $\alpha$ 1-fetoprotein, CD 68, cytokeratin, desmin and smooth muscle actin corresponded to bronchogenic cyst. The patient was discharged from the hospital on postoperative day 7; six months later on surgical check up the patient was well and symptom free. The patient has been continuously closely followed by a surgeon.

## Discussion

Bronchogenic cysts are rare congenital abnormalities with walls similar to those of normal bronchi and with pseudostratified columnar, ciliated, respiratory type epithelial lining and with bronchial wall components such as cartilage, smooth muscle, elastic fibers, fibrous tissue or seromucinous gland<sup>6</sup>. Sumiyoshi *et al.* have proposed a theory that the pathogenesis of bronchogenic cyst is caused by the pinching off of irregular lung budding of the primitive ventral foregut during the third to seventh weeks of intrauterine development. When attachment to the primitive foregut persists, the cyst is usually associated with the tracheobronchial tree or the esophagus. If complete separation occurs, the cyst may be found at other unusual locations<sup>7</sup>. Another theory that seems less probable is that such cysts represent aberrant differentiation of foregut-derived budding originating intra-abdominally<sup>8</sup>. In the retroperitoneal space, bronchogenic cysts are rare developmental anomalies. Reported cases occurred in both sexes at an equal ratio and over a wide age span ranging from 10 weeks to 59 years<sup>9</sup>. Most retroperitoneal bronchogenic cysts were found in the region of adrenal gland or adjacent to the stomach<sup>10-12</sup>. Retroperitoneal bronchogenic cysts are usually asymptomatic, secondary infection or compression of adjacent vital structures may become prominent and may induce clinical symptoms<sup>13,14</sup>. We found seven retroperitoneal bronchogenic cysts in association with the pancreas reported in the English literature. In three cases the location was the superior border of the pancreas body, in two cases they were related to the anterior

or of the pancreas, and in the remaining cases the cysts were adjacent to the superior border of the pancreatic tail and near the pancreatic neck. There were four female and three male patients, and the cysts measured between 2 cm and 7 cm in largest diameter<sup>12,13,15</sup>. Preoperative diagnosis based on clinical presentation and radiological appearances of bronchogenic cysts in the peripancreatic region may easily be misdiagnosed as pancreatic pseudocysts or pancreatic cystic tumors<sup>16-18</sup>. The histopathologic differential diagnosis of retroperitoneal cysts lined with respiratory epithelium includes cystic teratoma and bronchopulmonary sequestration. Cystic teratoma has endoderm-origin bronchial tissue but also other structures originating from the mesoderm and ectoderm. Bronchopulmonary sequestration can be diagnosed by the fact that it possesses lung parenchyma, pleural investment and bronchial elements. Cysts of mesonephritic and müllerian origin as well as esophageal cysts can also be covered by ciliated epithelium and thus misdiagnosed as bronchogenic cysts. The main difference from bronchogenic cyst is the absence of seromucinous glands or cartilage, and in case of esophageal cyst the presence of two smooth muscle layers in the cyst wall<sup>19,20</sup>. Complete surgical removal of the cyst is recommended to allow for accurate diagnosis and prevent complications. The most common complications are recurrence and secondary infection but malignant transformation to squamous and large cell carcinoma as well as adenocarcinoma has also been described<sup>21-23</sup>. Resection of previously infected cysts is more complicated and connected with a higher postoperative risk for the patient, therefore early removal of asymptomatic cysts is suggested. In conclusion, peripancreatic bronchogenic cysts, although extremely rare, should be considered on the differential diagnosis of cystic peripancreatic lesions. The difficulty in radiological preoperative diagnosis to exclude this type of cystic lesions from proliferative cystic tumors of the pancreas and possible complications justifies surgical approach.

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#### Sažetak

### RETROPERITONEALNA PERIPANKREATIČNA BRONHOGENA CISTA DIFERENCIJALNO DIJAGNOSTIČKI NALIK CISTIČNOM TUMORU GUŠTERAČE

M. Vučić, D. Tomas, M. Zovak, A. Mijić i B. Krušlin

Retroperitonealni cistični tumori su rijetki, a među njima bronhogeni retroperitonealni cistični tumori su iznimno rijetki. Bronhogene ciste su razvojne anomalije primitivnog crijeva koje najčešće nastaju u području prsne šupljine. Opisan je slučaj izolirane peripankreatične bronhogene ciste u 53-godišnjeg bolesnika koji je došao s anamnezom povremene epigastrične boli. Ultrazvuk abdomena i kompjutorizirana tomografija prikazali su cističnu leziju smještenu neposredno uz gornji rub repa gušterače. Postavljena je sumnja na cistični tumor gušterače. Cistična lezija je kirurški odstranjena u cijelosti, te poslana na patohistološku analizu. Mikroskopski cistu je oblagao pseudostrotificirani, cilindrični, cilijarni tip respiracijskog epitela sa žarišnim vrčastim stanicama. Stijenka ciste bila je vezivno-mišićna uz prisutne seromukozne žlijezde i žarišne upalne infiltrate, što je odgovaralo dijagnozi bronhogene ciste. Iako je rijetka u ovom području, bronhogeni cistu treba razmotriti u diferencijalnoj dijagnozi peripankreatičnih cističnih lezija.

**Ključne riječi:** *Bronhogeni cista – komplikacije; Bronhogeni cista – dijagnostika; Neoplazme gušterače – patologija; Neoplazme gušterače – dijagnostika; Retroperitonealne neoplazme – dijagnostika; Retroperitonealne neoplazme – patologija; Retroperitonealne neoplazme – kirurgija*