Case Report

Urologia Internationalis

Urol Int 2000;64:216–219

Received: January 6, 1999
Accepted after revision: January 21, 2000

Bronchogenic Cyst

A Rare Differential Diagnosis of Retroperitoneal Tumors

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Key Words

Cysts · Bronchogenic cyst · Retroperitoneal space pathology · Retroperitoneal space surgery · Retroperitoneal neoplasm surgery · Respiratory tract diseases · Neonatal diseases and abnormalities

Abstract

Subdiaphragmatic bronchogenic cysts are rare, and those located retroperitoneally are exceptional. A review of the English-language literature revealed only 16 reported cases. We describe an additional case of a retroperitoneal cyst presenting as an asymptomatic adrenal mass and discuss clinical, radiographic, surgical and pathological findings as well as its embryological background.

Introduction

Bronchogenic cysts are foregut-derived developmental abnormalities of the respiratory tract most commonly encountered in the mediastinum and rarely in the abdomen. Complete separation from the lung bud can give rise to unusual locations: bronchogenic cysts have been found attached to the sternum [1], pericardium [2], skin [3], spinal cord [4] and within the diaphragm [5]. Only a few cases of bronchogenic cysts occurring below the diaphragm are known from the literature, they have mostly been found intra-abdominally attached to the gastrointestinal tract, however, their occurrence in the retroperitoneum is extremely rare. We present clinical, radiographic, surgical, and pathological findings of a case of retroperitoneal bronchogenic cyst in a supra-adrenal location in an asymptomatic, adult patient.

Case Report

A 46-year-old man presented with 8 weeks' history of pain in the right thigh radiating to the knee. Neurological-neurosurgical work-up revealed a herniated disc at the L4/5 level. Magnetic resonance imaging that led to the above-mentioned, clinically suspected diagnosis also demonstrated a cystic mass of 3.8 × 3.5 cm situated in the right retroperitoneum. It was interpreted as a probable adrenal tumor.

On admission for surgery, his blood pressure was 130/75 mm Hg; serum potassium level of 4.2 mmol/l, and serum calcium level 2.4 mmol/l. He had a diffuse goitre I° together with mild tachycardia (110 bpm) and because of a slightly elevated T_3 level (3.3 nmol/l) and suppressed TSH, the patient was put on perchlorate (300 mg/day). To rule out metastatic disease, we performed a chest X-ray and ultrasound of the abdomen and testes, which were all negative. Urine dopamine levels were found to be slightly elevated (3,800 and 4,200 nmol/24 h). However, an iodine–131-MIBG assay (metaiodobenzylguanidine) did not demonstrate an adrenal or distant pheochromocytoma.

An exploration was made through a transperitoneal approach, in the position suggested on magnetic resonance imaging examination, and a fluctuating cystic mass was encountered. It was excised while the intact adrenal gland was left in place.

Macroscopically, the specimen appeared unilocular with a smooth, grey-brown outer surface and contained about 50 ml of yellow mucinous fluid. Microscopically, the inner wall was wholly lined by pseudostratified, columnar, ciliated epithelium with a thickened basement membrane. Exterior to the latter, sparse bundles of smooth muscle fibers, partly ossified hyaline cartilage and seromucinous glands, all identical to those found in the bronchus, were present. These findings confirmed the cyst as bronchogenic.

The patient had an uneventful recovery and was discharged on the 10th postoperative day.

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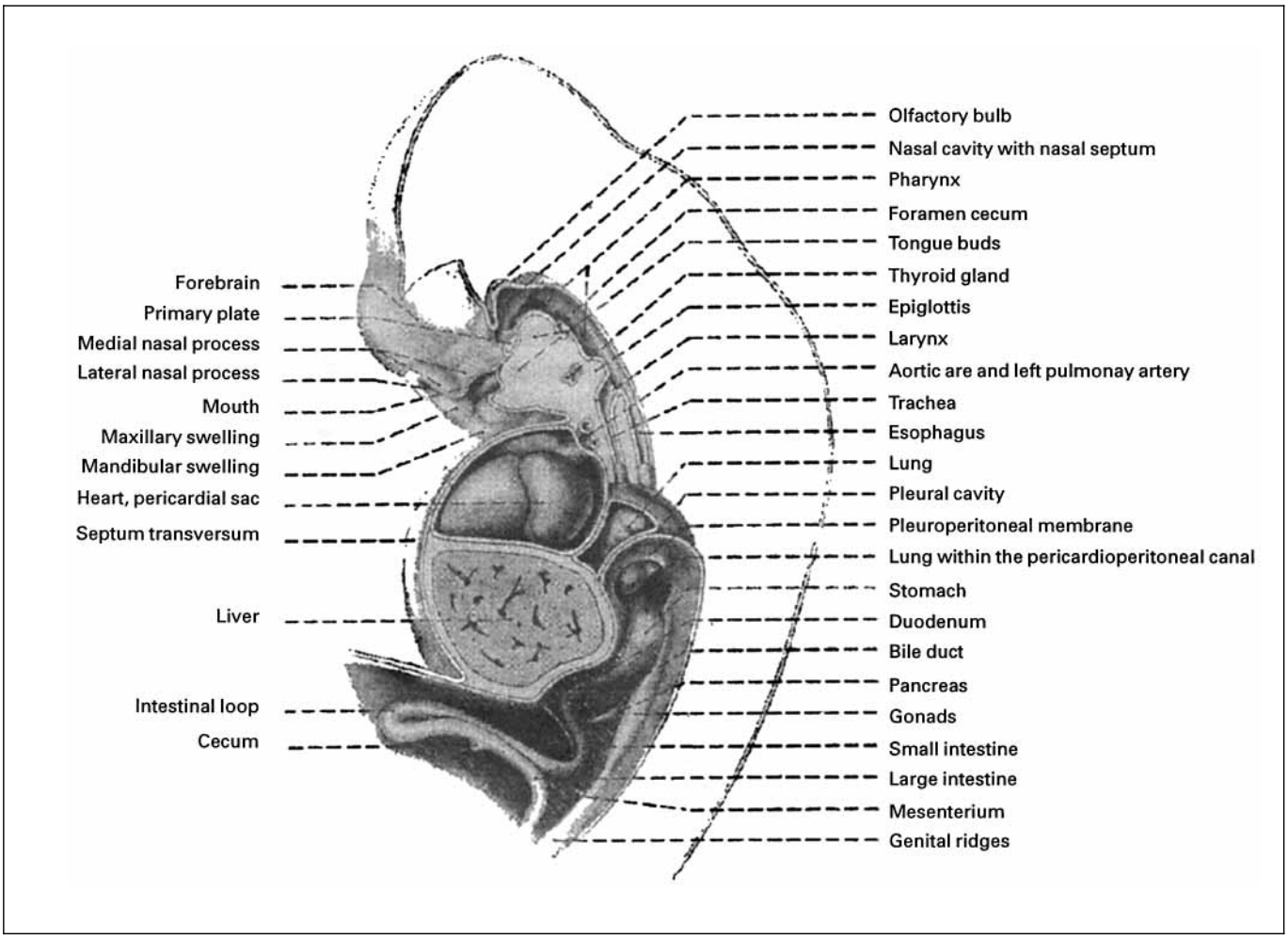


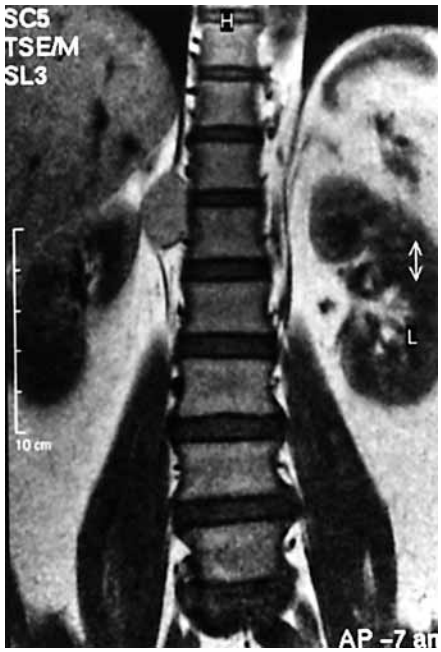
Fig. 1. Human embryo, C-R length: 12 mm, 6th week (modified from Benninghof [16]).

Comment

Since the first reported case of a bronchogenic cyst by Meyer [12] in 1859, several reports and reviews have appeared in the literature [13–15, 19–25]. If they are localized above the diaphragm, cough, dyspnea, pain and pneumonia may characterize the clinical presentation, whereas gastrointestinal symptoms are likely in the case of an intra-abdominally localized cyst. Three of 7 patients with retroperitoneally situated cysts were symptomatic probably because they were found in the vicinity of the kidney or pancreas. There are 2 cases of incidentally discovered, asymptomatic bronchogenic cysts in the retroperitoneum: Swanson et al. [7] described a case of a 4-year-old Black girl originally presenting with an afebrile bacterial urinary tract infection; Wirbel et al. [10] discussed a patient who was admitted for right hernia repair. In both cases, probable adrenal tumors have been suspected by routine abdominal ultrasound.

Histology

The histological findings are typical of bronchogenic cysts, that is, respiratory epithelium, smooth muscle, cartilage, mixed serous and mucinous glands. The differential diagnosis of a retroperitoneal cyst lined with pseudostratified, columnar, ciliated epithelium includes teratoma, bronchopulmonary sequestration, and cysts of urothelial origin in addition to bronchogenic cyst. It seems to be justified to exclude a teratoma showing respiratory epithelium as the only histological feature. Bronchopulmonary sequestrations, although believed to share a common embryologic origin with bronchogenic cysts, possess lung



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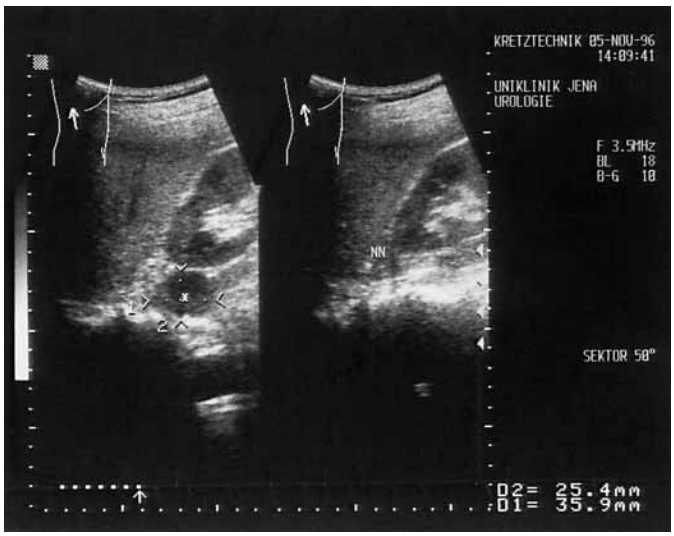

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Fig. 2. MRI scan showing a cystic mass adjacent to the right adrenal gland and spine.

Fig. 3. Ultrasound scans showing a 25 × 35 mm solid-appearing mass (left) that can be separated from the adrenal gland ('NN', right).

parenchyma and pleural elements. Neither the presence of respiratory epithelium nor the above-mentioned glands are seen in urothelial cysts.

Embryology

Bronchogenic cysts develop as a result of abnormal budding and pinching off of the tracheobronchial tree at about the 5th week of gestation. Although the derivatives are usually located in proximity to the bronchus and trachea, they may migrate to an aberrant location if connection with the tracheobronchial tree is lost. A possible explanation for retroperitoneally found bronchogenic cysts is as follows: the thoracic and abdominal cavity are linked by the pericardioperitoneal canal (fig. 1).

Prior to the fusion of the pleuroperitoneal membranes which subsequently results in forming of two respective cavities (divided by the diaphragm), abnormal buds of the tracheobronchial tree migrate into the abdomen. A less probable alternative explanation is the possibility of aberrant differentiation of a foregut-derived cyst originating intra-abdominally [8].

Radiographic Findings

As pointed out earlier, the diagnosis of a bronchogenic cyst was made incidentally: The patient was found at MRI to have a herniated disc explaining his 8-week history of radiating leg pain. As an additional finding, a cystic mass between the right adrenal gland and spine was seen. It appeared well defined and clearly demarcated from the surrounding structures indicating its benign nature (fig. 2).

Simple cysts normally show long T1 and long T2 relaxation times (signal intensity equal to or greater than skeletal muscle on T1 and brighter than fat on T2). In this case, we observed shorter T1 relaxation times – as did others [9] – which is probably due to the higher protein content of bronchogenic cysts [7]. We did not use gadolinium-containing contrast media, however, in the literature several authors report that bronchogenic cysts do not enhance [7, 9, 17]. Ultrasound scans show homogenous, solid-appearing masses often interpreted as adrenal tumors (fig. 3); however, the typical renal respiratory excursions of adrenal tumors were not seen in our patient. This might be a useful sonographic sign to differentiate tumors in that region.

Clinical Aspects

The differential diagnosis of retroperitoneal tumors (without adrenal tumors) and cysts is shown in tables 1 and 2, respectively. Once a solid mass of the adrenal region is suspected, the diagnostic work-up includes ultrasound, computed tomography and possibly MRI. To rule out hormone producing adrenal tumors appropriate chemical analyses, a careful history and accurate physical

Table 1. Primary retroperitoneal tumors (modified from Lehr et al. [18])

Mesenchymal tumors	50–70%
Malignant lymphoma	20–30%
Teratoma	5%
Ectopic epithelial tumors	5–10%
Neurogenic tumors	2–3% (higher in children)

Table 2. Retroperitoneal cysts

Mesenteric cysts
Mullerian duct cysts
Omental cysts
Lymphangiomatous cysts
Retroperitoneally localized meningocele
Echinococcus cyst
Giant hydronephrosis
Renal cyst

examination should be done. In particular, lung and testes are locations that need to be screened for primaries.

We preferred a transperitoneal approach offering the ability for complete abdominal exploration in case of adrenal metastasis including lymphadenectomy and generally recommend an exploration in case of incidentaloma > 3 cm since only surgical treatment leads to a definitive diagnosis besides the fact that these tumors/cysts may even become symptomatic at higher age [8,15].

In the meantime, sufficient experience has been achieved with retroperitoneoscopy at our institution. Considering the less aggressive approach and short hospital stay, we recommend this technique for small and mid-sized hormonally inactive incidentaloma.

In summary, we presented a rare case of a retroperitoneally localized bronchogenic cyst as an unusual differential diagnosis of retroperitoneal tumors. Patients meeting the typical imaging criteria and clinical findings of an asymptomatic bronchogenic cyst do not need surgical treatment.

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