

# A retroperitoneal bronchogenic cyst: a rare cause of a mass in the adrenal region

W J Haddadin, R Reid, R M Jindal

## Abstract

**This report documents a bronchogenic cyst presenting as an adrenal tumour in a 51 year old man with persistent epigastric pain. The cyst is regarded as a developmental abnormality of the primitive foregut, which typically occurs in the chest. Subdiaphragmatic, and retroperitoneal locations in particular, are unusual. The differential diagnosis of a bronchogenic cyst in the retroperitoneum includes cystic teratoma, bronchopulmonary sequestration, cysts of urothelial and mullerian origin, and other foregut cysts.** (J Clin Pathol 2001;54:801-802)

Keywords: bronchogenic cyst; retroperitoneum; adrenal; differential diagnosis

Primary retroperitoneal cysts are rare. Most originate from vestiges of embryonic blastemas. Their inner lining is usually compatible with either a mesothelial or mesonephric origin, although in some instances the lining is of mullerian type, with either a serous or mucinous appearance.<sup>1</sup>

Bronchogenic cysts are developmental abnormalities of the primitive foregut that are usually found above the diaphragm, especially in the mediastinum and particularly posterior to the carina. Rarely, they can occur in a subdiaphragmatic location, and a retroperitoneal position is distinctly unusual. To the best of our knowledge, there have been only 21 cases reported in the world literature, 15 of which have been in the English language. We report an isolated bronchogenic cyst in a 51 year old man that presented as an adrenal mass.

## Case report

A 51 year old white male patient, in whom a left hemicolectomy had been performed in December 1999 for a benign sigmoid colonic stricture secondary to complicated diverticular disease, presented approximately one month later with continuing epigastric pain, which he had been suffering from for the past 18 months. He had a past medical history of hypertension and reflux oesophagitis.

A computed tomogram (CT) of the abdomen was carried out, and showed a very well defined and circumscribed 5 cm solid mass in the left suprarenal region, displacing and compressing the left adrenal gland laterally (fig 1). The radiological appearances suggested the presence of a simple adrenal adenoma. A subsequent barium enema was carried out and revealed a reduced bowel calibre and associated spasm in the region of the previous anastomosis only. Routine laboratory studies were

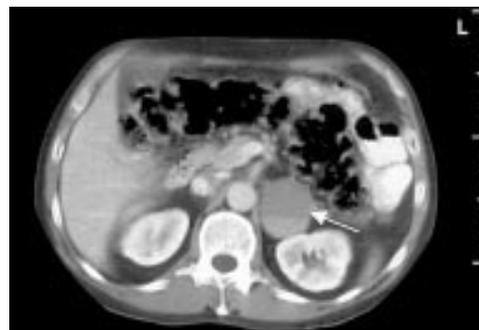


Figure 1 Computed tomographic scan showing a 5 cm solid mass in the left suprarenal region (arrow) compressing the adrenal gland laterally.

normal, including full blood count, urea, and electrolytes. Plasma cortisol and successive urinary catecholamine estimations were in the normal range.

In April 2000, the patient was admitted for surgical removal of the mass. At surgery, the left suprarenal region was explored through a left flank incision, taking out the 12th rib. A 5 cm cystic mass was discovered adherent to the upper pole of a normal looking left kidney. The cyst was dissected from the kidney and the left adrenal was found to be adherent to the supralateral part of the cyst. The entire cyst and the adrenal gland were excised. The cyst was opened on the theatre table. It contained thick whitish mucoid fluid. A sample of fluid was sent for bacteriological cultures, which were later found to be negative for microorganisms, including alcohol and acid fast bacilli. The postoperative course was uneventful and the patient was discharged four days later. On follow up, the patient's epigastric pain has subsided.

## Histopathology

Gross examination of the specimen showed a collapsed, previously opened, dark grey to brown cyst measuring 40 × 35 mm, with a maximum wall thickness of 3 mm. The inner lining was mostly smooth. The outer surface of the cyst showed an attached, partly fragmented and compressed, adrenal gland measuring 30 × 25 × 3 mm, with adjacent small amounts of fat tissue. The entire cyst was sectioned and submitted for microscopic examination.

Histologically, the cyst was found to be lined by respiratory-type, pseudostratified, ciliated columnar epithelium, resting on fibrous connective tissue focally containing seromucous glands and nodules of hyaline cartilages, fulfilling the histological criteria of a bronchogenic cyst (fig 2). There was no cytological atypia in the lining epithelium or stromal tissue components. Neither pulmonary parenchyma nor teratomatous

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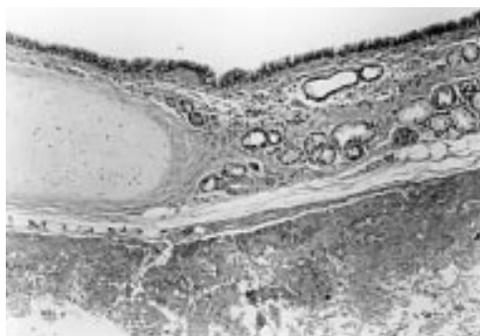


Figure 2 The cyst lining composed of respiratory-type epithelium, underlying lamina propria containing cartilage, and mixed mucous and serous glands (haematoxylin and eosin; original magnification,  $\times 400$ ).

components could be identified. The outermost part of the cyst showed attached adrenal gland with normal cortex and medulla.

### Discussion

Bronchogenic cysts are thought to be foregut malformations.<sup>2</sup> They arise from an abnormal budding of the tracheobronchial anlage of the primitive foregut during the 3rd to 7th weeks of development. When attachment to the primitive foregut persists, the cyst is usually associated with the tracheobronchial tree or the oesophagus. If complete separation occurs, the cyst may occur in other unusual locations, presumably by migration.<sup>3</sup> A retroperitoneal location is exceptionally unusual. Although the exact mechanism is unknown, Sumiyoshi *et al* proposed the following theory: in early embryonic life, the thoracic and abdominal cavities are linked via the pericardio-peritoneal canal. When the canal is later divided by fusion of the pleuroperitoneal membranes (the future diaphragm), a portion of the tracheobronchial tree could be pinched off and could migrate, resulting in a retroperitoneal bronchogenic cyst.<sup>4</sup>

Retroperitoneal bronchogenic cysts have been reported to occur in both sexes in equal ratio, and in a wide age range. Among the 21 cases reported in the world literature, the oldest patient was a 59 year old man,<sup>4</sup> and the youngest was a 3 month old boy,<sup>5</sup> in whom a subdiaphragmatic cyst was diagnosed prenatally at 25 weeks of gestation on routine scan.

Most retroperitoneal bronchogenic cysts have been found in the region of the left adrenal gland or the superior body of pancreas. These cysts are generally too small to cause symptoms—most reported cases measured less than 5 cm in diameter and were picked up incidentally. Among patients who presented with symptoms, most complained of epigastric or back pain; their cysts measured over 7 cm in diameter, with the largest reported case being 10 cm in greatest dimension.<sup>6</sup> Interestingly, that case was reported in a 44 year old man who presented with phaeochromocytoma-like symptoms attributed to compression of the adrenal gland by the cyst with secondary release of catecholamine by the gland.

In our case, the cyst was diagnosed after evaluation of a patient for persistent epigastric pain. The cyst measured 5 cm in diameter and

was found in the left suprarenal region compressing the adrenal gland. There was no evidence of endocrine abnormality.

Based on clinical and radiological appearances, a retroperitoneal bronchogenic cyst is easily misdiagnosed as an adrenal cortical or medullary tumour, pancreatic cyst, enteric cyst, cyst of urothelial origin, etc. However, a histological diagnosis of a bronchogenic cyst from these lesions can be made.

The differential diagnosis of a retroperitoneal cyst lined by pseudostratified ciliated columnar epithelium includes cystic teratoma, bronchopulmonary sequestration, cysts of urothelial and mullerian origin, and other foregut cysts, in addition to a bronchogenic cyst.<sup>7</sup> In our case, a teratoma was excluded by the absence of tissue representing the three different germinal layers. A bronchopulmonary sequestration possesses lung parenchyma and a pleural investment, along with bronchial elements, which were absent here. The presence of subepithelial cartilage and seromucous glands ruled out the possibility of mullerian and urothelial cysts. Among foregut cysts, those containing cartilage or seromucous glands are classified as bronchogenic cysts, as in our case; those containing two well developed layers of smooth muscle without cartilage are classified as oesophageal cysts; and those with none of these distinguishing features are classified as foregut cysts of gastric, enteric, or combined gastroenteric types.<sup>2</sup>

The treatment of retroperitoneal bronchogenic cyst is surgical removal. Although most are asymptomatic, excision is recommended to establish diagnosis, alleviate any symptoms, and prevent complications, especially infections and the remote but documented risk of malignant transformation.<sup>8</sup>

This unusual case supports the previously published reports of retroperitoneal bronchogenic cysts, and further emphasises the difficulty of preoperative diagnosis, because CT images may suggest the presence of a solid mass, probably because the cyst contains thick proteinaceous secretions.

In summary, although bronchogenic cysts are rare, they should be considered in the differential diagnosis of a retroperitoneal mass, particularly a cystic tumour in the region of the left adrenal gland.

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