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# Adrenal bronchogenic cyst masquerading as an incidentaloma: Case report and review of literature

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**Abstract**
Bronchogenic cysts are, rare developmental anomalies of ventral foregut, resulting from an aberrant budding of primitive tracheobronchial tree during third to seventh week of gestation. They are commonly found in thoracic cavity, in the region of posterior mediastinum and rarely at other sites like retroperitoneum. Retroperitoneal bronchogenic cysts involving adrenal gland is exceptionally rare with only fifty cases of adrenal bronchogenic cysts reported in literature. We present a case of a young 30 year old adult male who presented with left flank pain and right inguinal hernia. Contrast enhanced CT abdomen revealed a hypodense lesion in the region of left adrenal gland. The patient underwent open excision of tumor with Left adrenalectomy and was discharge on postoperative day five. Histopathologically, the tumor was seen to be lined with ciliated columnar epithelium suggestive of bronchogenic cyst. No complications or recurrence occurred after three years of follow up. Adrenal bronchogenic cysts are exceptionally rare tumors. It should be considered as differential diagnosis of Adrenal Incidentaloma.

**Keywords:** bronchogenic cyst, retroperitoneal, adrenal gland, incidentaloma

## Introduction
Bronchogenic cysts arise as an aberrant out pouching from foregut derivative, the tracheo-bronchial tree [1]. Having maintained the connection to the foregut derivatives, they are often located to the thoracic cavity, mostly posterior mediastinum. Rarely, losing their connection, they migrate to extra-thoracic sites, like skin, neck, retroperitoneaum and peritoneal cavities. Retroperitoneal bronchogenic cysts, as first reported by Miller in 1953 are rare [2]. Adrenal involvement has been reported in fifty cases of retroperitoneal bronchogenic cysts [3]. Most remain asymptomatic, unless the size increases or complications like, cyst infection, haemorrhage or malignant transformation ensue. Preoperative diagnosis is exceptional as there are no established diagnostic criteria and often the diagnosis can only be confirmed by histopathology. Histopathologically bronchogenic cysts are defined by the presence of-stratified; ciliated columnar epithelium accompanied by anyone of the following-Cartilage, smooth muscle or seromucus glands [2, 4]. We aim to report a case of left adrenal gland bronchogenic cyst and review its radiological, clinical characteristics and management.

## Case Report
A young 30 year old adult male, presented to us with left flank pain and right inguinal hernia. While investigating, ultrasound abdomen revealed a 5 cm, hypoechoic lesion in the region of left adrenal gland. Contrast enhanced CT abdomen revealed a 5 cm, hypodense lesion in the region of left adrenal gland (Figure 1). The investigations to know the functionality of the tumour were proven to be negative. Specific tumor markers including carcinoembryonic antigen (CEA) and carbohydrate antigen 19-9 (CA19-9) were normal. The left adrenal gland was approached by left, subcostal, transperitoneal incision. The tumour along with left adrenal gland could easily be excised except for the superior aspect, where a tubular structure of sizeable diameter was seen to enter the gland. The structure was clamped and cut. The tumour was cut open to reveal chocolate coloured mucinous secretion (Figure 2). Histopathologically, the tumour was seen to be lined with ciliated columnar epithelium along with islands of hyaline cartilage and seromucus glands, thus fulfilling the defining criteria (Figure 3). Postoperative period was uneventful. No complications or recurrence after three years of follow up.

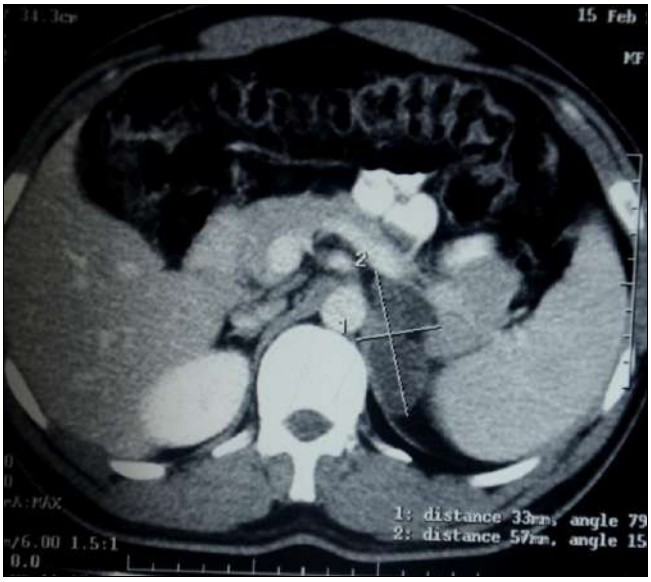


**Fig 1:** CT Scan picture showing a hypo dense lesion in the region of left adrenal gland.

**Fig 2:** Left Adrenal cut section

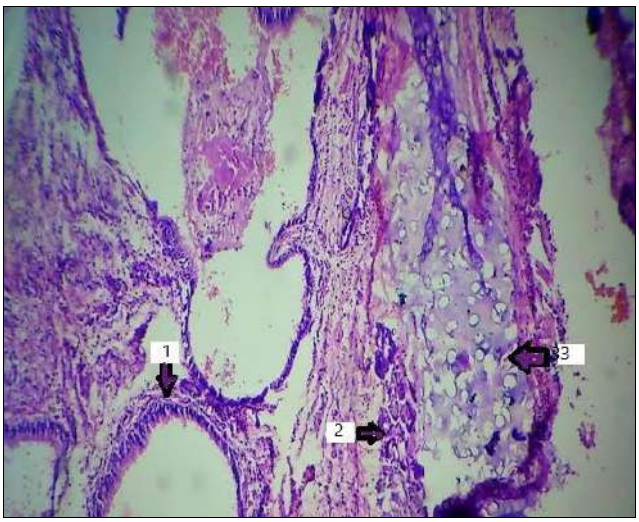


**Fig 3:** Photomicrograph showing Pseudostratified ciliated columnar epithelium (1); Hyaline cartilage (2); Seromucus glands (3) [40X magnification; H&E stain]

## Discussion and Review

Miller *et al*. were the first to report retroperitoneal bronchogenic cysts. In the past confusion about histopathological criteria to diagnose bronchogenic cysts, has resulted in over reporting of cases in the literature. Kim Coverts *et al*. [2] in 2012 did a Medline search and found reports of 62 cases of retroperitoneal bronchogenic cysts out of which, after applying strict criteria of presence of Stratified, ciliated epithelium along with cartilage or smooth muscle or seromucus glands, only 30 cases were true retroperitoneal bronchogenic cysts. Similarly review by Xiao J *et al*. [3] in 2022 using Pubmed database included 107 cases in literature with their own series of 19 cases showing true bronchogenic cyst. Our case met all the histopathological criteria and hence is a true retroperitoneal bronchogenic cyst.

Bronchogenic cysts are the out pouching of tracheo-bronchial tree, in great majority of cases maintain their connection with the parent structures and hence are found in thoracic cavity in the region of Posterior mediastinum (50%); Superior mediastinum (14%) and Pericarinal area (35%) [5]. Rarely they lose their attachments and migrate to other areas like retroperitoneum. The cysts reported in literature are more likely to be detected in left abdomen (76.6%), upper abdomen (94.6%) and retroperitoneal space (76.6%) with the three most frequent areas for were the left adrenal region (36.0%), pancreatic region (10.8%), and gastric cardia/lesser curvature of the stomach (8.1%) [3]. Several hypothesis have been put forth to explain the sub diaphragmatic location of bronchogenic cysts, the one proposed by Sumiyoshi *et al*. in 1985 seems to be most plausible [6]. According to their theory, the aberrant buds are pinched off the tracheo-bronchial tree by the developing diaphragm. Since the left pleuroperitoneal canal is larger of the two and closes late, these cysts are said to be preponderant on the left side. They are said to predominate in a triangular area bordered by midline, splenic vein & diaphragm [7] and left adrenal gland is said to be the commonest site of retroperitoneal bronchogenic cysts. In the 126 cases of true bronchogenic cysts as reported by Xiao J *et al*., left adrenal gland was the site of cyst in 47 cases. Bilateral adrenal Bronchogenic cyst has been reported in one case [8].

The retroperitoneal bronchogenic cysts occur in both males and females in equal measure and show a wide variation in age at presentation, the youngest being a child of 3 months. The size at presentation varies from 3 cm to 18 cm but most reported cysts have been less than 5 cm in diameter.

Bronchogenic cysts are mostly asymptomatic, but as they are lined by a secretory epithelium, tend to increase in size over a period of time and become symptomatic. As they increase in size, can compress the adjacent structures. Anderson *et al*. [9] reported a case of retroperitoneal bronchogenic cyst, mimicking phaeochromoctoma due compression of adjacent adrenal gland. The cysts can get infected and develop acute haemorrhage as well. Malignant transformation of bronchogenic cysts is rare and most of the cases reported, the cysts were in thoracic cavity. Only one case of retroperitoneal bronchogenic cyst developing adenocarcinoma has been reported in the literature [10].

A confident preoperative radiological diagnosis is exceptional, as there are no established diagnostic features. On CT abdomen, these may occur either as hypodense [0-20 HU] or mixed density lesions, sometimes accompanied by

punctuate calcifications or milk of calcium. Hyper attenuation [up to 120 HU] indicates intracystic haemorrhage or presence of thick mucus or proteinaceous material [11]**.** MRI shows the cysts to be of variable intensity and offer no additional diagnostic help over CT abdomen.

Treatment consists of surgical excision either by open surgical method or laparoscopic or retroperitoneaoscopic methods, the latter two offering an advantage of being less morbid with promise of earlier return to activity for the patients. The indications for surgery of these largely asymptomatic, limited size lesions consist of establishing the diagnosis, relieve symptoms and to prevent any potential malignancy risk [12].

Conclusion**:** Adrenal bronchogenic cysts are exceptionally rare tumours, which mimic other adrenal tumours and form an important differential diagnosis of Adrenal incidentalomas and retroperitoneal neoplasm. Long term results of adrenal bronchogenic cyst are excellent with no recurrence after surgical excision.

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**Conflict of Interest:** None Declared

**Consent:** A written consent was taken from the patient to publish clinical details & relevant photographs

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