



## RETROPERITONEAL BRONCHOGENIC CYST- (AN UN-COMMON ENTITY AT AN UNCOMMON SITE.)

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**ABSTRACT:-** Bronchogenic cysts can occur rarely in a sub diaphragmatic location, as a part of the morphologic spectrum of bronchopulmonary foregut malformations.

They usually present due to compression of nearby structures or found incidentally. Here we report case of retroperitoneal bronchogenic cyst mimicking adrenal gland tumor. To date, few cases of retroperitoneal bronchogenic cyst has been reported in the English literature. One of the earliest reports was in 1953, by Miller et al.

**Keywords:-** Adrenal mass, Bronchogenic cyst, Embryology, Pseudostratifiedcolumnar epithelium Retroperitoneal ,

### I. INTRODUCTION

Bronchogenic cysts are rare congenital anomalies of primitive foregut that usually develop in the posterior mediastinum, most along the tracheobronchial tree.<sup>(1)</sup> Retroperitoneal locations have been described earlier still it is a extremely rare.<sup>(2)</sup>

These patients are commonly asymptomatic and their diagnosis is made incidentally.<sup>(3)</sup> It has been suggested that these tumors grow, bleed, or compress adjacent structures, thereby becoming symptomatic. Infections and cystic perforations have been described.<sup>(4)</sup>

### II. CASE HISTORY

A 28 yrs male patient underwent examination for non specific chronic abdominal pain.

**2.1 USG Abdomen:** Showed a well-defined hypo echoic focal lesion in left suprarenal region? Adrenal mass.

**2.2 CT Abdomen:** Showed heterogeneous enhancing lesion in left adrenal region –likely? Adenoma/ mass.

Because a definitive diagnosis was uncertain, the patient underwent surgical excision of the lesion. Excised specimen was sent for histopathology examination.

The patient had an uneventful post operative recovery.

#### 2.3 HISTOPATHOLOGY:

**2.3.1 Gross:** Fig. 1. Specimen consisted of hemorrhagic multi cystic mass measuring 4.5x3x3cm .The cysts were of varying sizes with largest measuring 3x3cm and contained mucoid material and outer surface of cyst showed an attached and compressed adrenal gland measuring 3.7x1.2x0.5cm .Entire cyst was sectioned and submitted for microscopic examination.

#### 2.3.2 Microscopy:

Multiple section studied showed cysts of varying sizes lined by pseudo stratified ciliated columnar epithelium with cyst filled with hemorrhagic and mucoid fluid, with areas of haemorrhage in between the cysts. Some of the cysts showed underlying hyaline cartilage and smooth muscle.

Section studied from adrenal gland was unremarkable.

Features suggested: Retroperitoneal Bronchogenic cyst of adrenal gland.

Fig. 2. (Hematoxylin and eosin 4x)The cystic lining is composed of a thin layer of epithelium (arrowhead) covering loose connective tissue and some bronchial glands (arrow).

Fig. 3. (Hematoxylin and eosin 40 x ) detail of the cystic wall with respiratory-type ciliated columnar epithelium (arrowhead), containing scattered smooth muscle fibers.

Fig. 4. (Hematoxylin and eosin 4x) showing hyaline cartilage and pseustratified ciliated columnar epithelium.

Fig. 5. (Hematoxylin and eosin 40x) showing pseudostratified ciliated columnar epithelium and hyaline cartilage.

### III. DISCUSSION

The first report of a retro peritoneal bronchogenic cyst was described in 1953 by Miller et al.<sup>(5)</sup> These tumors are located commonly in thoracic cavity and retroperitoneal site is extremely rare.

Bronchogenic cyst is typically unilocular and arises in any one of the three mediastinal compartments, neck or retro peritoneum. Bronchogenic cysts arise from an abnormal budding of the tracheobronchial analog of the primitive foregut.

When the attachment persists, the cyst is usually found near or associated with the tracheobronchial tree or the esophagus. If a complete separation occurs, the cyst may migrate to other unusual locations. The exact mechanism of its migration is still unknown.

Sumiyoshi et al<sup>(6)</sup> hypothesized that the thoracic and abdominal cavities are linked by the pericardioperitoneal canal in an early embryonic stage. When the canal is divided into respective cavities by fusion of the pleuroperitoneal membranes (the future component of the diaphragm) at the end of the 6th week of intrauterine life, abnormal buds of the tracheobronchial tree are pinched off by these membranes and migrate into the abdomen, resulting in a retroperitoneal bronchogenic cyst.

Histological characteristics of typical bronchogenic cyst include ciliated respiratory type epithelium on the surface and smooth muscle, nodules of hyaline cartilage in the wall and seromucinous glands.

To detect a bronchogenic cyst, CT scan and MRI are the best imaging modalities. In the CT scan the lesions are mixed cystic-solid, sharply defined, hypoattenuating and homogenous possibly with calcifications.

In T2-weighted MRI the cyst shows an extremely bright signal. These have been the primary imaging modalities for evaluating and defining the nature of these masses and their anatomic relations.

It has been reported that bronchogenic cysts occur in both sexes in equal ratio, and in a wide age range.

In 1997, Hsieh et al reported the first case of retroperitoneal bronchogenic cyst in Taiwan.

Current English literature, a total of 44 cases of isolated retroperitoneal bronchogenic cysts has now been reported<sup>(7)</sup>. Accordingly, the most common locations are as follows: (1) superior left adrenal gland (43%); (2) left crus of the diaphragm (16%); (3) superior pancreas tail (12%) ;( 4) right crus of the diaphragm (11%); (5) superior pancreas body (7%). The reason for this phenomenon is still unknown, but its presume that it is related to the pathway of embryologic development.

A systematic review of PubMed had been performed by Olof jannasch et al<sup>(8)</sup> using the following search strategy: bronchogenic cyst AND (adrenal OR retroperitoneal OR sub diaphragmatic). 18 BC being removed via minimally invasive approaches had been found. Till now 19 cases were reported out of which 18 were located left-sided. Ours is also left sided. . Including our own case 8 cases were removed retroperitoneoscopically and 12 laparoscopically<sup>(9)</sup>

**This table shows details of articles concerning with periadrenal bronchogenic cysts.**

Publication (year)	Age	Gender	Location	Side	Size(cm)	Surgical approach	Epithelium	Cartilage	Muscle	Glands	Hd
<b>Mccrystal dj (2002)2 cases</b>	8	Female	Adrenal	Left	4*3*2	Retrop.	Resp., duct-like structures	yes	Yes	Seromucinous	Bc
	15	male	Adrenal	Left	5.5*3.5*1.2	Retrop	Lined cystic	yes	Yes	Seromucinous	Bc
<b>Ishikawa t (2003)</b>	41	Female	Adrenal	Left	5.5*5.2*9.2	Retrop.	Tall columnar	yes	Yes	Seromucinous	Bc
<b>Chung m(2009)</b>	41	Female	Adrenal	Left	4.8*3.5*4.2	Retrop.	Psce	yes	Yes	Seromucinous	Bc
<b>O'neal pb(2012)</b>	23	Female	Adrenal	Left	5.2*4	Retrop.	Psce	yes		Bronchial mucous	Bc
<b>Jannasch o(2013)</b>	50	Male	Adrenal	Left	2.7*2.7*5	Retrop.	Psce	yes		Seromucinous	Bc

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<b>Alguraanz(2012)</b>	23	Female	Adrenal	Right	N.r	Lap	N.r	n.r		N.r	Bc
<b>Akosmb92006)(hungarian)</b>	18	Female	Supra adrenal	Left	6*8	Lap	Multilayer, ciliated, columnar	no		Mucous	Bc
<b>Chupy(2007)</b>	55	Male	Adrenal	Left	4*3	Lap	Psce	yes		no	Bc
<b>Mineis(2007)</b>	39	Male	Adrenal	Left	4.5	Lap	Psce	yes		mucous	Bc
<b>Terryne(2007)</b>	75	Female	Lateral of adrenal	Left	5	Lap	Ciliated	yes		Yes	Bc
<b>Obandoj(2009)</b>	67	Male	Adrenal	Left	3.9*3.7	Lap	Ciliated tufts	yes		Bronchial mucinous	Bc
<b>El youssefer(2010)</b>	44	Male	Adrenal	Left	3.1*1.6	Lap	Ciliated columnar	n.r		Glandular	Bc
<b>Rud o(2010)(german)</b>	51	Female	Adrenal	Left	3.5*2.5*2.5	Lap	Ciliated	yes	Yes	Bronchial mucinous	Bc
<b>Powellg(2012)</b>	50	Male	Peri adrenal	Left	2.3	Lap	Psce, cuboidal	yes	Yes	Seromucinous	Bc
<b>Cai y(2012)</b>	50	Female	Adrenal	Left	3	Lap	Psce	yes	Yes	Seromucinous	Bc
<b>Tokudan(1997)</b>	24	Female	Adrenal	Left	3	Lap	Psce	n.r	N.r	N.r	Bc
<b>Ishizuka o(2004)</b>	36	Male	Adrenal	Left	5*3	Lap	Psce	n.r	N.r	N.r	Bc
<b>Roma a(2008)</b>	40	Male	Adrenal	Left	6.2	Lap	N.r	n.r	N.r	N.r	Bc
<b>Ours</b>	28	Male	Supra adrenal	Left	4.5*3*3	Retrop.	Psce	yes	Yes	Bronchial mucous	Bc

**Lap**-laparoscopic; **retrop**-retroperitoneoscopic; **n.r**-not reported; **BC**-bronchogenic cyst; **resp**-respiratory; **PSCE**-pseudostratified ciliated epithelium; **HD**-histological diagnosis.

Laboratory abnormalities were uncommon in patients with retroperitoneal bronchogenic cyst. There were some patients who had their metanephrine and catecholamine analyzed, which showed mild elevations. This phenomenon might be attributable to compression of the adrenal gland by the cysts, with secondary release of catecholamine from the gland. In our case urine VMA levels were mildly elevated (112.2mmol/24hr) with mild lower levels of cortisone hormone (57.10nmol/L).

Malignancy arising in bronchogenic cysts has been reported only rarely, and most occur in the mediastinum as squamous cell carcinoma, fibrosarcoma, undifferentiated carcinoma, leiomyosarcoma, embryonal rhabdomyosarcoma, and anaplastic carcinoma. So far, only 1 case of a retroperitoneal bronchogenic cyst attached to the ascending colon with associated adenocarcinoma has been reported.<sup>(9)</sup>

The treatment of retroperitoneal bronchogenic cyst is surgical removal via laparoscopy or laparotomy.

#### IV. CONCLUSION

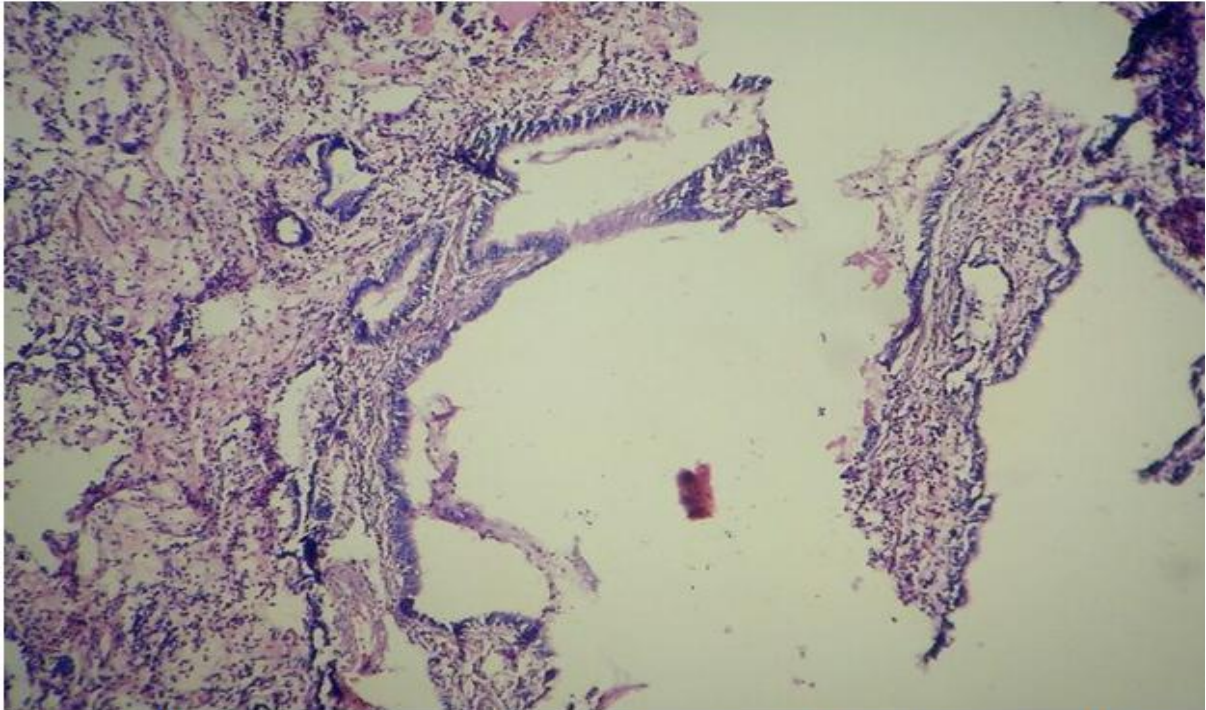
We diagnosed a rare case of bronchogenic cyst in a 28 year old male, who had the complaints of left flank pain. Bronchogenic cyst must be considered in differential diagnosis of cystic tumors located in retroperitoneal region. Despite the progress in imaging modalities, surgical resection is mandatory to achieve a definitive diagnosis.<sup>(10)</sup> These cysts can be differentiated from other lesions only by **histopathological examination**. Accurate diagnosis is important to rule out malignancy in order to avoid aggressive surgery.

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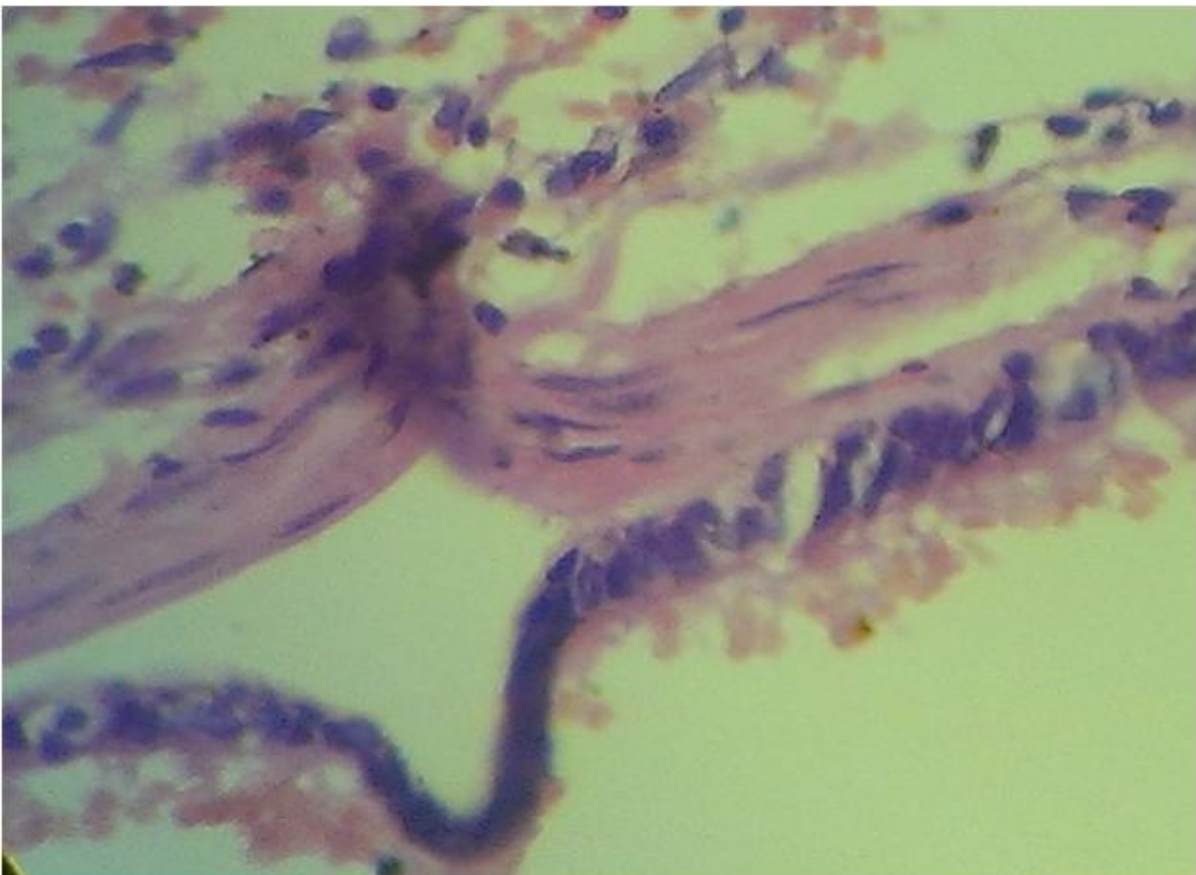
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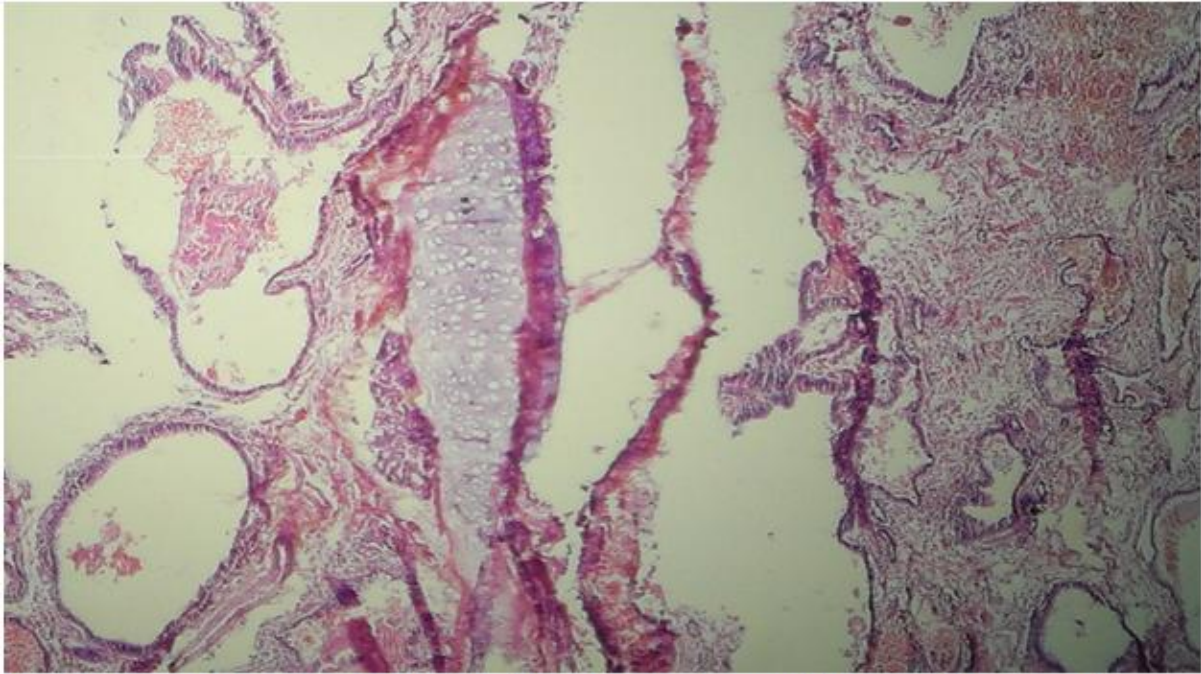
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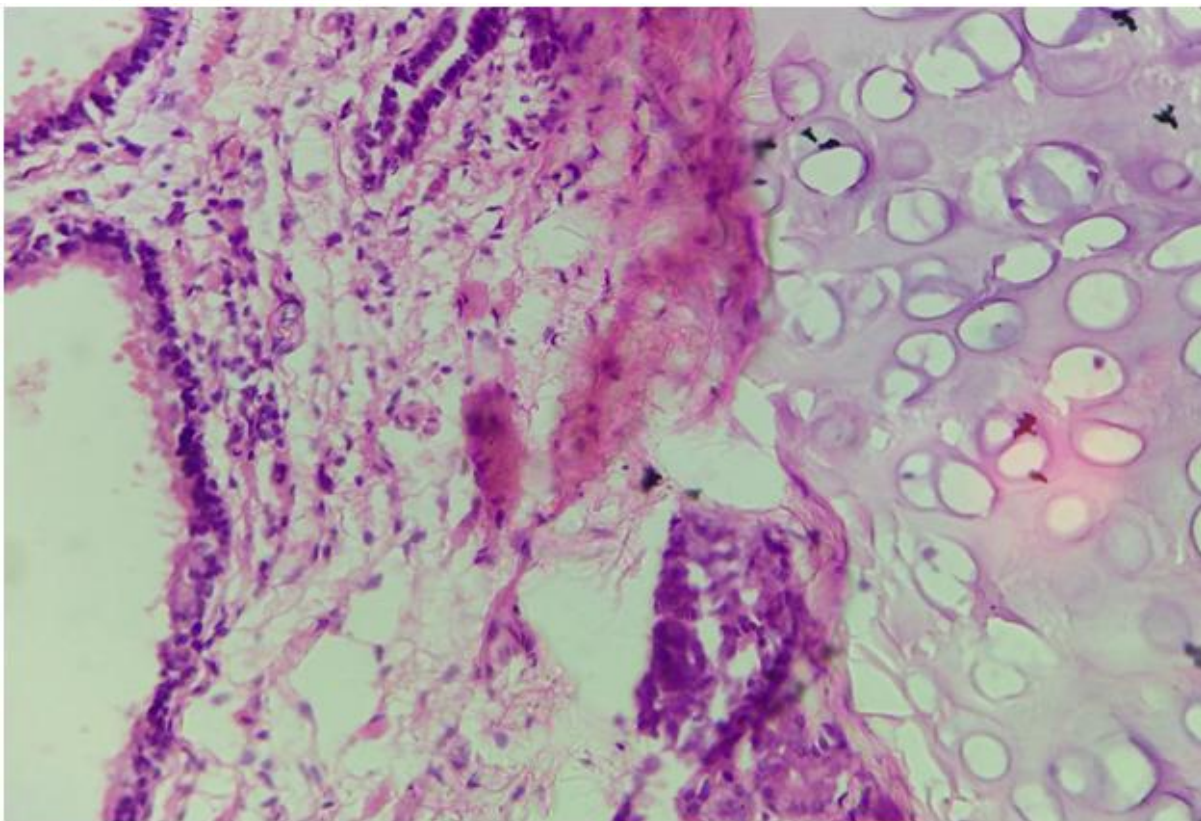
**Fig. 2. (Hematoxylin and eosin 4x)**The cystic lining is composed of a thin layer of epithelium (arrowhead) covering loose connective tissue and some bronchial glands (arrow).



**Fig. 3. (Hematoxylin and eosin 40 x )** detail of the cystic wall with respiratory-type ciliated columnar epithelium (arrowhead), containing scattered smooth muscle fibers.



**Fig. 4. (Hematoxylin and eosin 4x) showing hyaline cartilage and pseudostratified ciliated columnar epithelium.**



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