

Case Report

Challenge in preoperative diagnosis of retroperitoneal mucinous cyst in a pediatric patient

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Abstract: Mucinous cystic lesions of the retroperitoneum can be either neoplastic or non-neoplastic. It is very important to make a correct diagnosis, or at least, an accurate classification, to proceed with an optimal treatment strategy. In spite of advantage of ultrasound and X-ray image examinations, it is still a challenge to make differential diagnosis of retroperitoneal mucinous cyst from gangliocytoma because both tumors have similar density under the image assessment. In this article, we reported an asymptomatic 8-year-old boy with multiple bronchogenic cysts in both lung and adrenal area on the left side, the latter was considered to be a gangliocytoma preoperatively by ultrasound and computed tomography, but confirmed as bronchogenic cyst by histopathology post laparoscopic resection. The differential diagnosis, imaging features and treatment of bronchogenic cyst are discussed and the relative literatures are reviewed.

Keywords: Retroperitoneal mass, mucinous cyst, bronchogenic cyst, child

Introduction

Because therapeutic strategies for retroperitoneal cystic masses vary depending on the cause, the ability to noninvasively differentiate between masses is very important. Despite the progress made in imaging diagnosis methods, such as ultrasound, computed tomography (CT) or magnetic resonance imaging (MRI), which can provide important information regarding mass location, shape, size and blood supply, the presence and thickness of a wall, the presence calcifications or fat, involvement of adjacent structures [1], it is still hard to make an accurate diagnosis of mucous cyst in retroperitoneal region. Here we report a pediatric patient with retroperitoneal cyst, which diagnosed as ganglioneuroma preoperatively, while the final diagnosis of bronchogenic cyst (BC) was made according to histopathology after laparoscopic resection.

BC is regarded as a benign, congenital developmental abnormality, which develops from

abnormal budding or branching of the tracheo-bronchial tree during embryogenesis [2]. It is rather uncommon, Coselli reported that it accounts for 1/42,000 and 1/68,000 admissions in two hospital series [3]. Most BCs develop in the thorax within the mediastinum or in the lung parenchyma [2], in rare cases, it can occur anywhere along the developmental pathway of the foregut due to mis-migration [4]. Ectopic locations other than thorax were reported, such as diaphragm [5, 6], esophagus [7, 8], stomach [9], ileal mesentery [10], neck [11, 12], abdominal wall [13], periscapular area [14], and so on. Most cysts measured <5 cm in diameter exhibit no symptoms, which are usually related to secondary infection or compression of adjacent structures. The most common complaint of BC is epigastric, flank or back pain. Although it is a cyst, BC can cause severe complications in some cases [15], it also has malignant potential [16-18]. Fortunately, according to the literature, the prognosis of most BCs is excellent after complete excision.

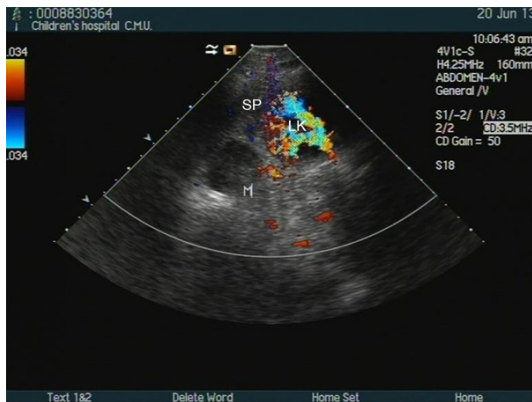


Figure 1. Abdominal ultrasound imaging of the mass in left adrenal region, about 36 mm×34 mm×31 mm, with low density echo, mucoid composition, no obvious blood fluid signal inside the mass (M: mass; LK: left kidney; SP: spleen).

Case history

An 8-year-old boy without any symptoms was referred to our hospital because of a mass in the left adrenal region which was incidentally discovered by ultrasonic examination during routine check-up. He had no hypertension and other significant medical history, and no remarkable family history. Results of physical examination were unremarkable. Laboratory tests including blood cell count, renal function, electrolytes, cortisol, renin, aldosterone, and adrenocorticotrophic hormone (ACTH) were in the normal range. Serum alpha fetoprotein (AFP) and urine vanillyl mandelic acid (VMA) were negative.

Abdominal ultrasound (SIEMENS Sequoia512) revealed a mass in left adrenal region, about 36 mm×34 mm×31 mm, with low density echo, mucoid-like contents, no obvious blood fluid signal inside the mass (**Figure 1**). No remarkable findings on chest radiograph. CT (GE light Speed VCT 64 Slice) of abdomen scan demonstrated a 39 mm×26 mm ovoid-shaped homogeneous mass located in the left retroperitoneal region, about 10 Hounsfield units, and the mass was enhanced slightly, about 17 Hounsfield units (**Figure 2**), no other abnormalities found in the abdomen, such as enlargement of lymph node or ascitic fluid, indicating a benign mass. Radiological appearances suggested the presence of a retroperitoneal ganglioneuroma or cyst. 3 pulmonary cystic masses were observed in chest CT, 11 mm~16 mm

in diameter, congenital pulmonary cyst was diagnosed.

Since tumor mass could not be excluded, laparoscopic resection of the retroperitoneal lesion was performed. On operation, a cystic mass was excised entirely, which was founded located between left kidney and adrenal gland, about 40 mm in diameter, with 2-4 mm thick wall, adhered with the surrounding tissues, seriflux inside the mass and ruptured during operation, semitransparent egg-white-like liquid was observed flowing out, no cartilage or hair was found. The left kidney and adrenal gland were normal. The patient recovered well postoperative and discharged 6 days later.

Histopathology examination showed that the inner side of cyst was lined with ciliated columnar epithelium (**Figure 3**), surrounded by consecutive smooth muscle layer, local fibrous connective tissue contained seromucous glands, infiltrated with chronic inflammatory cells. No cartilage was observed. The final pathology diagnosis was left retroperitoneal BC.

The parents refused further operation on pulmonary BC due to no symptom and rare chance of malignant transformation, the patient is now under close following up. The boy was followed up for 21 months postoperative so far, he was in good condition, no reoccurrence and other abnormal found by abdominal ultrasound examination, no obvious enlarge of the pulmonary cysts.

Discussion

Retroperitoneal mucinous cystic mass is not very common, which could be neoplastic lesions, such as cystic teratoma, cystic lymphangioma, mucinous cystadenoma, cystic mesothelioma, cystic change in solid neoplasms, pseudomyxoma, or nonneoplastic lesions such as adrenal or pancreatic hematoma [19], epidermoid cyst, tailgut cyst, pancreatic nonneoplastic cyst, urogenital cyst [20, 21], and bronchogenic cyst. Taken advantage of imaging techniques, such as ultrasound, CT or MRI, it is still a great challenge to make an accurate diagnosis preoperatively, because the thick proteinaceous secretion may masquerade characteristics of solid masses, such as neuroblastoma, adrenal adenoma, pheochromocytoma, pan-

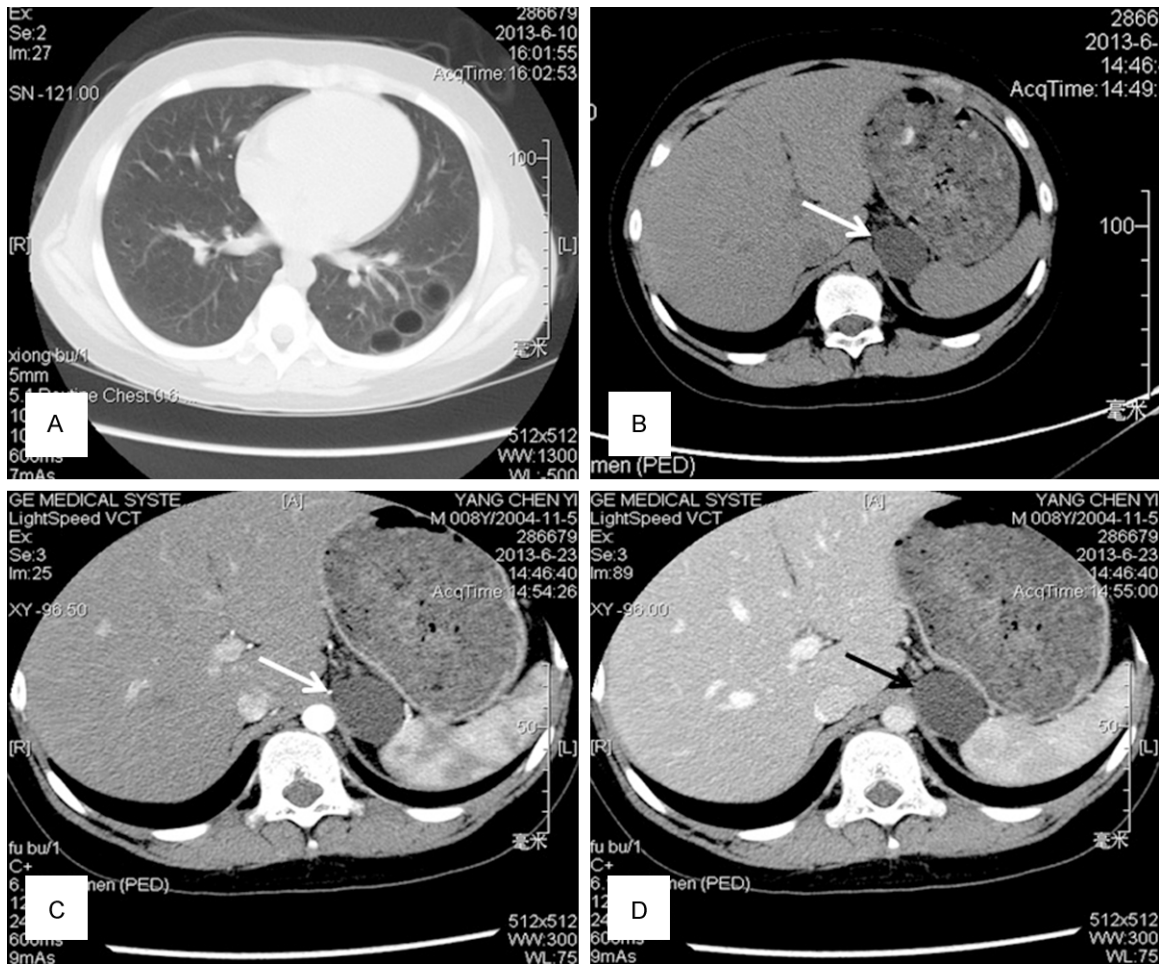


Figure 2. Computed tomography scan of chest and abdomen. 3 pulmonary cystic masses were observed in chest, 11 mm~16 mm in diameter (A). A 39 mm×26 mm ovoid-shaped homogeneous mass located at the left retroperitoneal region, about 10 Hounsfield units (B), and the mass was enhanced slightly, about 17 Hounsfield units in arterial phase (C) and venous phase (D).

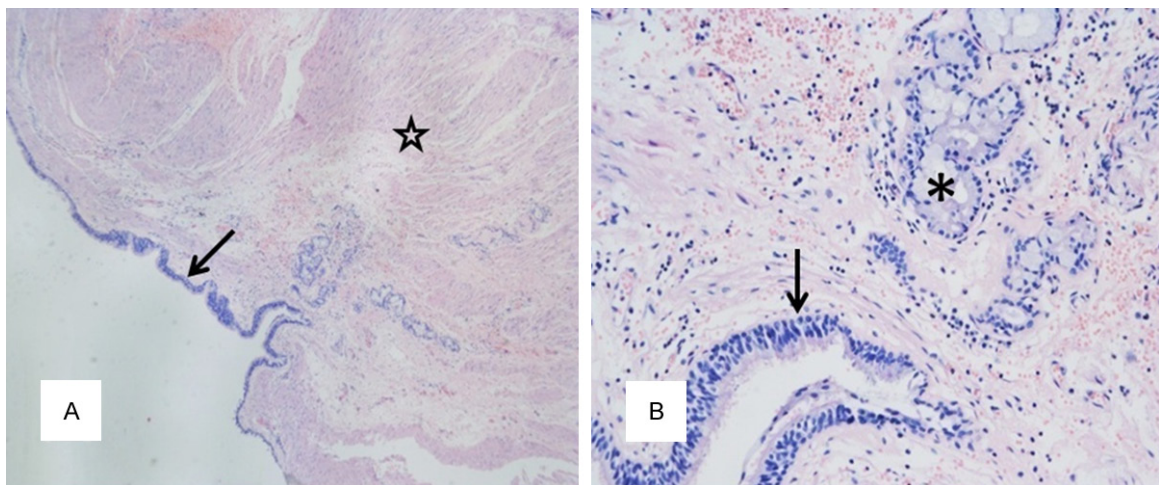


Figure 3. Histopathology examination showed that the inner side of cyst was lined with ciliated columnar epithelium (arrow), surrounded by consecutive smooth muscle layer (star), local fibrous connective tissue contained seromucous glands (*), infiltrated with chronic inflammatory cells (HE stain; A: ×40; B: ×200).

Retroperitoneal mucinous cyst in children

Table 1. Pediatric patients with extra-thorax bronchogenic cysts reported in the literature

Age	Gender	Complaint	Location	Size of mass	Imaging manifestation	Treatment	Pre-operation diagnosis	History	Ref
Since birth	F	a small opening on the epigastrium	abdominal wall	20 mm×3 mm	US: a short hypoechoic tubular lesion extending from the external opening to the fatty subcutaneous layer of the abdominal wall	surgery resection	dermoid cyst, bronchogenic fistula, or a cutaneous cyst	-	[13]
3 d	F	midline cervical cleft	neck	3 mm×5 mm×2 mm	US: a non-specific area of cystic change inferior to the cleft in the midline, thyroid gland is normal	surgery resection	-	-	[12]
1 y	F	a dark-green drainage at the back of the shoulder	scapular skin	15 mm×11 mm	US: a lesion with a thick wall and a hypoechoic center, no connection with the thoracic cavity was seen	surgery resection	-	-	[14]
19 m	F	Fever of unknown reason	right hemidia-phragm	40 mm×30 mm	chest X-ray: A suspicious lung lesion on the right hemi-thorax; CT: A cystic lesion in the segment VIII of liver indicating a hydatid cyst	explorative lapa-rotomy	liver hydatid cyst	fever	[6]
20 m	F	-	abdomen	-	-	-	-	recurrent urinary tract infections	[31]
3.5 y	M	asymptomatic	omental bursa	30 mm in diameter	MRI: a simple cyst, located in the retrogastric region	open surgery resec-tion	-	constipation	[32]
4 y	F	-	gastroesophage-al junction area	20-30 mm (CT)	US: a left upper pole simple renal cyst and a solid left adrenal mass CT: a left renal cyst and a 2-3 cm soft tissue mass of 30 HU MRI: adrenal mass with a long T2 and a neutral T1 signal	open surgery resec-tion	-	UTI, urinary incontinence	[33]
8 y	F	central abdominal pain associated with anorexia, nausea for 3 days	left adrenal gland region	50 mm×24 mm×20 mm	Abdominal radiograph: unremarkable US: normal apart from a quadrilateral-shaped mass measuring 40 mm×36 mm×25 mm with a well-defined capsule superior to the left kidney. The echogenicity was similar to that of the renal cortex and less than that of the adjacent spleen. Follow-up US 6 months later confirmed that the mass was enlarging CT: Replacement of the left adrenal gland by a rhomboid mass with uniform internal structure. Minimal enhance-ment. No calcification	excision via a poste-rior retroperitoneo-scopic approach	benign adrenal adenoma	-	[26]
10 y	M	-	paraesophageal intra-abdomen	34 mm×21 mm×25 mm (MRI)	US: A mass, considered as intrahepatic, hypoechogenic, with a 2.5 cm diameter CT: An intrahepatic mass, with sharp margins and at-tenuation, slightly higher than water, containing small calcifications. No enhancement MRI: An extrahepatic, well-circumscribed cyst, located in the lesser omentum, contacting liver and especially the left hepatic vein	laparoscopic resec-tion	hepatic tumor	-	[8]
12 y	F	severe abdominal pain	ileal mesentery	60 mm in diameter	US: A 6 cm right ovarian cyst	laparoscopic resec-tion	ovarian cystic mass	-	[10]
12 y	M	-	right retroperito-neal region	-	CT: small, hyperdense nonenhancing mass adjacent to the right crus of the diaphragm	open surgery resec-tion	-	neuroecto-dermal tumor of the pelvis	[34]
15 y	M	lower left posterior tho-racic pain for 6 months	left adrenal gland region	50 mm×48 mm×30 mm (CT)	CT: a calcified, well-defined mass in the left adrenal fossa, with no enhancement	excision via a poste-rior retroperitoneo-scopic approach	ganglioneuroma	-	[26]

US: Ultrasonography; CT: Computed tomography; MRI: Magnetic Resonance Imaging; d: days; m: months; y: years; M: male; F: female; UTI: urinary tract infection.

Retroperitoneal mucinous cyst in children

Table 2. The differential diagnosis of bronchogenic cyst with common retroperitoneal malignant tumors in children

Disease	Age	Symptom or sign	Imaging manifestation	Tumor marker	Ref
Neuroblastoma	median age: 21 months, <2 years: 50%, <4 years: 75%, <10 years: 95%	fever, anorexia, body weight loss, arthralgia, abdominal mass	Plain radiographs: calcified abdominal or posterior mediastinal mass (30%) CT: invasive suprarenal mass, stippled calcifications (85%); rim calcifications, the most specific pattern for neuroblastoma (29%) MRI: superior to CT in assessing tumor encroachment into the neural foramen and spinal canal CT and MRI: Lobulated heterogeneous enhancing mass with internal hemorrhage and necrosis, with aggressive features: invasion of adjacent organs, engulfment of surrounding vessels (e.g. celiac axis, superior mesenteric artery or aorta) and distant metastases (most commonly to the liver or bone)	VMA, HVA, N-myc	[35]
Wilm's tumor	young children (median age, 3.5 years), with more than 80% of cases occurring in those younger than 5 years	abdominal mass, abdominal pain 25%, hematuria 25%, hypertension 25%, hemihypertrophy 3%	US: renal origin mass with blood supply, solid nature, and margin is smooth and well-defined, heterogeneous with hypoechoic and anechoic areas due to liquid haemorrhage, necrosis or cysts, calcification is less common (9%), renal vein and inferior vena cava thrombosis (4%-10%) CT: confirm US findings, the tumour enhances less than the normal adjacent kidney and clearly visualized in contrast imaging MRI: low signal intensity on T1-weighted sequences and hypo- or isointensity on T2-weighted images	WT1	[36]
Teratoma	Any age, often found in babies at birth and in young children, <10 years: 50%	asymptom or abdominal/pelvic pain, nausea and vomiting	US: Cystic, solid or mixed echogenicity mass CT: Heterogeneous cystic mass with focal calcifications and fat, better than US at defining the teratomas extent to the surrounding organs and in evaluating the cyst wall MRI with coronal and sagittal scans is superior to ultrasound and CT for demonstrating the anatomical relationship with adjacent organs such as abdominal aorta or spinal cord and local tumour spread	AFP	[22, 37]
bronchogenic cyst	Any age	asymptom or back pain	US: cystic mass, with no blood fluid in the central area CT: Rounded, solitary, sharply-defined, well-circumscribed, low density, homogeneous mass. The density can vary from typical water density to high density MRI: dependent on the content of the cyst, serous: Very low signal intensity on T1-weighted images and of very bright signal intensity on T2-weighted images; proteinaceous material: high signal intensity on T1-weighted images	-	[2, 23, 25]

US: Ultrasonography; CT: Computed tomography; MRI: Magnetic Resonance Imaging; VMA: vanillylmandelic acid; HVA: homovanillic acid; WT1: Wilm's tumour tumor suppressor gene 1; AFP: alpha-fetoprotein.

creatic neoplasm, metastatic testicular teratoma [22].

BC, as the case reported in this article, is an uncommon congenital nonneoplastic anomaly, due to foregut mis-migration during embryonic development. It usually locates within the mediastinum and the lung [2], ectopic BC other than chest is rare, and always misdiagnosed with other retroperitoneal masses. To the best of our knowledge, it's the first case of simultaneous occurrence of bronchogenic cyst in pulmonary and retroperitoneal region.

Although BC is a congenital abnormality, it can occur at any age, range from newborn to over 70 years old [2, 13], always be discovered incidentally on routine check-up or for other non-related conditions. According to the literature, there were very few cases diagnosed as BC in extra-thorax region in children, and the locations were variety (**Table 1**). Due to the variety of age, location, and rare occurrence, BC shows a wide range of clinical and radiologic manifestations, which makes the correct preoperative diagnosis extremely difficult [23]. The most common retroperitoneal malignant tumors in children, neuroblastoma, wilm's tumor, and teratoma, which are easily misdiagnosed with BC, are listed and compared with BC in **Table 2**. Among all of the BCs occurred in the retroperitoneal region reported in the literature, more than 80% have been identified on the left side [24].

The useful imaging examinations of abdominal, retroperitoneal or chest mass include ultrasonography, CT, and MRI. On ultrasonography, BC is usually described as a cystic mass, with no blood supply in the central area. The typical appearance of BC on CT is a rounded, solitary, sharply-defined, well-circumscribed, low density, homogeneous mass; the shape may be irregular when compressed. The density can vary from typical water density to high density related to increased calcium content, anthracotic pigment, blood, or greater protein content of the fluid. No obvious enhancement in contrast-enhanced image [23]. The thick proteinaceous secretion, calcium or the anthracotic pigment contents of bronchogenic cysts may masquerade characteristics of solid masses which may then be mistaken for retroperitoneal tumor. MRI can be useful for elucidating the cystic nature of these lesions [25]. The MRI

appearance is dependent on the content of the cyst, if the fluid within a BC is serous, it will be of very low signal intensity on T1-weighted images and of very bright signal intensity on T2-weighted images. However, many BC may contain large amounts of proteinaceous material, which have a characteristic appearance with high signal intensity on T1-weighted images [2].

The definitive diagnosis of BC is established only by surgical excision and tissue histology examination. BC is typically lined with ciliated, pseudostratified, columnar epithelium with possible areas of malpighian metaplasia, the cyst wall contains airway components such as bronchial glands, cartilage plates, smooth muscle, and occasionally nerve and adipose tissues can also be observed [2, 26]. In some instances, BC with complex structure is thought to be teratoma. Roma et al. studied 22 BC and 34 retroperitoneal teratomas metastatic from testicular tumors, about 77% of BC showed tracheobronchial differentiation, whereas none of the teratomas did [22].

The operative resection of BC is controversial, because it's a benign lesion and usually asymptomatic. But, several complications of BC, including recurrence, ulceration, infection, hemorrhage, malignant transformation, and even lethal complications have been described [15-18], thus surgical excision of BC is recommended [27]. According to the literature, most surgeons adopted surgical removal to treat retroperitoneal BC, to establish the definite diagnosis, prevent potential complications or the risk of tumor formation, and to alleviate any symptoms, such as back or flank pain in some cases [23, 28]. Retroperitoneoscopic and laparoscopic operation are viable as a less aggressive approach with less invasive and shortened hospital stay [29]. Larger lesions or violations of the surrounding adjacent tissues may require to open surgery [28]. Complete excision of the whole cyst is essential, because any remnants of the cyst epithelial tissue can cause relapse. In this case, the retroperitoneal mass was supposed as ganglioneuroma preoperatively, and it was finally confirmed as BC by histological examination after laparoscopic resection of the retroperitoneal mass, while the asymptomatic small pulmonary cysts are still in closely followed up.

The prognosis of BC after complete excision is excellent in most of patients as reported in the literature. However, recurrence is possible, even 24 years post-operative as reported [30]. What's more, malignant transformation of BC was reported in several patients [16-18]. So, patients with BC need long-term following up in the life span.

In short, it's still a great challenge to make an accurate pre-operative diagnosis of retroperitoneal mucinous cystic mass, which could be either neoplastic or nonneoplastic lesions, what's more, there's risk of malignant transformation for uncommon nonneoplastic lesions, therefore, we recommend explorative surgery resection for uncertain retroperitoneal mucinous cystic mass.

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Disclosure of conflict of interest

None.

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