RESEARCH ARTICLE

RETROPERITONEAL BRONCHOCYSTIC CYST. A CASE REPORT

N. Hammoune1, M. Boui1, S. Belhind1, S. Belaasri1, A. El. Guezzar2, M. Azami3, M. Atmane1 and A. Mouhsine1

1. Department of Radiology, Avicenne Military Hospital, Marrakech, Morocco.
2. Department of Surgery, Avicenne Military Hospital, Marrakech, Morocco.
3. Department of Pathology, Avicenne Military Hospital, Marrakech, Morocco.

Abstract
Bronchogenic cyst is a rare congenital abnormality, most commonly found in the mediastinum. Retroperitoneal location presents only 0.03% of all sites. The exact etiopathogenesis is not yet well known, it seems to be a malformation arising from the primitive foregut. RBC is generally asymptomatic and often misdiagnosed, radiological imaging helps to find out the cyst and guide the diagnosis. The final diagnosis is made by histological analysis. We report a case of a 33-year-old patient with retroperitoneal bronchogenic cyst, located adjacent to the tail of pancreas and ileal loop, the imaging is misleading, reminiscent of a gastrointestinal stromal tumor, the mass was surgically completely removed, the unsuspected diagnosis was assessed by pathology.

Introduction:–
Bronchogenic cyst is a rare congenital abnormality, usually occurring in the mediastinum (1), close to the tracheobronchial tree, posterior to the carina. Retroperitoneal location presents only 0.03% of all sites (2). The exact etiopathogenesis is still poorly known, it would seem to be a malformation arising from the primitive foregut. Most of the patients with retroperitoneal bronchogenic cyst (RBC) are asymptomatic, when there are symptoms; they are related to the cyst development or complications.

Radiological imaging helps to find out the cyst and guide the diagnosis. Retroperitoneal bronchogenic cyst is often misdiagnosed as teratomas, lymphoceles, neoplasms derived from the pancreas or the adrenal system (3). The final diagnosis is confirmed by histological analysis. We report a case of a RBC of a 33-year-old man.

Case report:
A 33-year-old male without neither medical nor surgical history, was admitted to the hospital with abdominal pain of one year duration. Physical examination was normal as serum laboratory tests. A computed tomography (CT) of the abdomen was carried out and revealed an irregular-shaped mass located in the upper retroperitoneal space, measuring 102x68x108mm (anterior-posterior axis x width x height) it seemed to have a double component; fluid and tissular (Fig1). In addition, calcifications were present at the periphery of the cystic zone (Fig1). The lesion was located adjacent to the tail of the pancreas and ileal loop. Subsequently enhanced computed tomography demonstrated a poor enhancement of the mass and no evident enlarged lymph nodes in the retroperitoneal space. (Fig 2)
Based on these findings, some diagnoses were suggested like; gastrointestinal stromal tumor (GIST) or neoplasms derived from the pancreas. Therefore, we completed the imaging by an abdominal CT scan with oral contrast to increase the location accuracy and try to specify if the mass was at the expense of ileal loop. (Fig 3) The lesion was surgically completely removed via midline laparotomy using a left retroperitoneal approach, the tumor was not adherent to any other structures and was resected without difficulty (Fig 4) (fig5) On gross examination of the specimen showed a multilocular cystic mass splited by some fibrous septas of varying thickness. (Fig 6) The middle and high magnification ( haematoxyline and eosin stain) of a histological section showed a cystic lesion bordered by a pseudostratified ciliated respiratory-like epithelium supported on a wall composed of smooth muscle cells associated with mucus glands. (Fig7). A diagnosis of retroperitoneal bronchogenic cyst was made. The postoperative course of the patient was uneventful.

Discussion:
Retroperitoneal bronchogenic cysts are known as benign congenital cysts, supposed to be the result of an abnormal budding of the tracheobronchial anlage of the primitive foregut during the third and seventh week of gestation.

Most bronchogenic cysts are localized in the mediastinum or lung parenchyma, and other locations are rare. Most of the sub diaphragmatic bronchogenic cysts are located in the peritoneal cavity and are attached to the gastrointestinal tract or communicate with it. Retroperitoneal locations are extremely rare, accounting for only 0.03 % of all tumors. Embryologically, the exact mechanism is still unclear. Sumiyoshi and Al assumed that; in early embryonic life, the thoracic and abdominal cavities are linked via pericardio-peritoneal canal. When the canal is later divided by fusion of the pleuropertitoneal membranes, a part of the tracheobronchial tree could be pinched off in the abdominal cavity, resulting in retroperitoneal bronchogenic cyst (4). Clinically, retroperitoneal bronchogenic cysts are usually asymptomatic, unless they are infected or large enough to cause local compression of adjacent organ. Most common symptoms are vague abdominal discomfort, early satiety and lower back pain (5).

Sonography even though restricted by the deep retroperitoneal location and gastrointestinal gas, could confirm the cystic nature of the lesion: indicating an anechoic lesion with or without echogenic debris (6). The CT images typically demonstrate a thin-walled, well-defined, lobulated cystic mass, generally filled with water-density content, without enhancement. It could be hyperdense due to proteinaceous, thick mucinous or hemorrhagic content (6,7). Some cysts show calcification and milk of calcium. At MRI, variable signal intensity has been described. Most of the cysts are slightly hyperintense or isointense to skeletal muscle on T1-weighted images and hyperintense on T2-weighted images. The signal intensity of the cyst contents is not suppressed on T1-weighted fat suppressed images. On gadolinium enhanced T1-weighted images, there is an enhancement of the cyst wall, aiding the delineation of the thin wall. In the current case MRI and sonography exams have not been performed. The CT scan showed a mixed mass located in the upper retroperitoneal space sharply defined, heterogenous suggesting tissular lesion with a cystic area (6,7,8). Based on clinical and radiological appearances, the lesion was misdiagnosed as a possible gastrointestinal stromal tumors or cystic lymphangioma. The retroperitoneal bronchogenic cyst diagnosis had not been suggested.

Due to the embryological development in the foregut, RBC are composed of mesodermal and endodermal tissue. On pathology they are lined by ciliated pseudostratified columnar epithelium that rests on a connective tissue wall and may have focal areas of hyaline cartilage, bronchial glands, and smooth muscles in their walls similar to the bronchial wall.

These features were found in the presented case. Although bronchogenic cysts are almost invariably benign, complications such as inflammation, infection, and compression of adjacent structures have been described. In our case the benign character of the mass had been supposed first of all; clinically due to chronic and poor symptoms without deterioration of general condition, then the radiological imaging did not showed any invasion sign or enlarged lymph nodes.

Surgical treatment is indicated to confirm the diagnosis and to prevent potential complications.

Bronchogenic cysts located in the retroperitoneum can be excised using either a laparotomy incision or a flank incision. The prognosis is excellent. (9,10)
Figure 1: Axial abdominoplevic unenhanced computed tomography (CT) showing an irregularly-shaped, mixed solid cyst mass (white arrow) in the left retroperitoneal region, with peripheral calcifications of some cystic zones (star).

Figure 2: Axial abdominoplevic enhanced computed tomography (CT) showing a poor enhancement of the mass.

Figure 3: The lesion do not become opacified on abdominal CT scan with oral contrast.
Figure 4: Left retroperitoneal surgical approach indicating that the tumor was not adherent to any other structures.

Figure 5: Surgical specimen of the mass.
Figure 6: Voluminous multilocular cystic mass split by some fibrous septas of varying thickness.

Figure 7: Haematoxyline and eosin stain photomicrograoh (middle and high magnification) of the histological section showed a cystic lesion bordered by a pseudostratified ciliated respiratory-like epithelium (white arrow) supported on a wall composed of smooth muscle cells associated with mucus glands.

Conclusion:
A bronchogenic retroperitoneal cyst, although rare, should be included in differential diagnosis of a retroperitoneal mass.

Conflicts of Interest:
The authors declare that there are no conflicts of interest regarding the publication of this paper.

Reference: