Sub-Diaphragmatic Bronchogenic Cysts: Report of Two Cases

Sub-Diyafragmatik Bronkojenik Kist: 2 Olgu Sunumu

Intraabdominal Bbronchogenic Cyst

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Özet

İntra-abdominal yerleşimli bronkojenik kistler son derece nadirdir. Çoğunlukla intratorasik yerleşimli olmalarına rağmen, diyafragma altında da nadiren yerleşim gösterirler. Bu çalışmada diyafragma altında saptanan ve cerrahi eksizyon yapılan 2 olgu literatür bilgileri ışığında sunuldu.

Anahtar Kelimeler

Bronkojenik Kist; Cerrahi Tedavi; Ekstratorasik; Konjenital

Abstract

An intra-abdominal tumor originating from a bronchogenic cyst is an extremely rare entity. Although commonly located in the thoracic cavity, these cysts are occasionally present underneath and even within the diaphragm. We report two cases of bronchogenic cysts localized in the extra-thoracic region and treated with surgical therapy, in light of the current literature.

Keywords

Bronchogenic Cyst; Congenital; Extra-Thoracic; Surgical Treatment

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Introduction

Bronchogenic cysts are rare congenital developmental anomalies of the foregut. They are usually localized in the mediastinum. Rarely, bronchogenic cysts are reported in extra-thoracic regions such as the sub-diaphragmatic area or cervical region [1-3]. When these cysts are localized in an intra-abdominal region, their radiological and clinical diagnosis is difficult because they may resemble gastrointestinal stromal tumors, neuroendocrine tumors, leiomyoma, and pancreatic pseudocysts [4-6]. In this study, we will present two cases of sub-diaphragmatic bronchogenic cysts treated with surgical resection.

Case Report 1

A 35-year-old woman presented with epigastric pain during menstruation periods for 1 year. Clinical examination revealed epigastric tenderness; her laboratory tests were within normal limits except for a mild increase in CA 125 with a level of 64.97 U/ml (reference range 0-35 U/ml). An ultrasound scan and computed tomography of the abdomen revealed a tubular thin walled cystic lesion, approximately 8x7x4 mm, connected to the antrum of the stomach, right lobe lateral segment of the liver and the corpus of the pancreas with nodular components. (Figure 1A). She underwent an operation via laparotomy with the pre-diagnosis of mesenteric cyst, pancreatic cyst, or duplication cyst. At operation, the cystic lesion was identified and excision was performed totally with its capsule. Histological examination showed an 8.5x5x5 cystic lesion with 2-3 mm of wall thickness and a mucoid content. The cyst was lined ciliated columnar respiratory epithelium containing smooth muscle tissue and sero-mucinous acini on the wall. The pathological diagnosis was a bronchogenic cyst.

Case Report 2

A 54-year-old male patient was admitted to the hospital with chest pain. Computed tomography revealed a regular bordered, oval lesion of 9x7x5cm in diameter on the postero-medial part of the diaphragm medial to the spleen, surrounded by calcifications (Figure 1B). Left posterolateral thoraco-phrenotomy was performed. Exploration found a regular bordered, encapsulated lesion with thick wall that was attached to the peritoneal surface of the diaphragm in a small region, lying through the left crus of the diaphragm without any invasion of the surrounding structures, and containing gelatinous fluid. The lesion was explored thoroughly. The pathology report was a cystic lesion lined with stratified ciliated epithelium containing a bronchogenic wall with sero-mucinous glands surrounded by cartilage areas. These findings were compatible with bronchogenic cyst (Figure 2A,B).

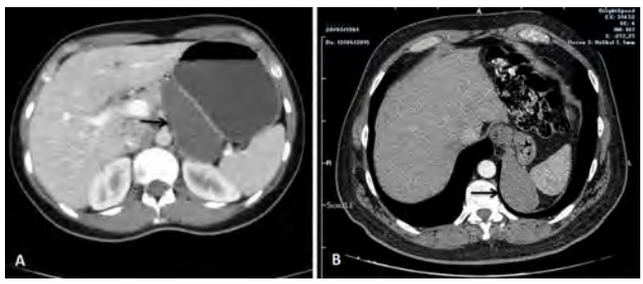


Figure 1. Findings from computed tomography - a 9.5x4cm tumor lesion of the posterior side of antrum of the stomach (A). Computed tomographic scan showing a 9x7x5 cm in diameter regular bordered mass on the left postero-medial part of the diaphragm (B).

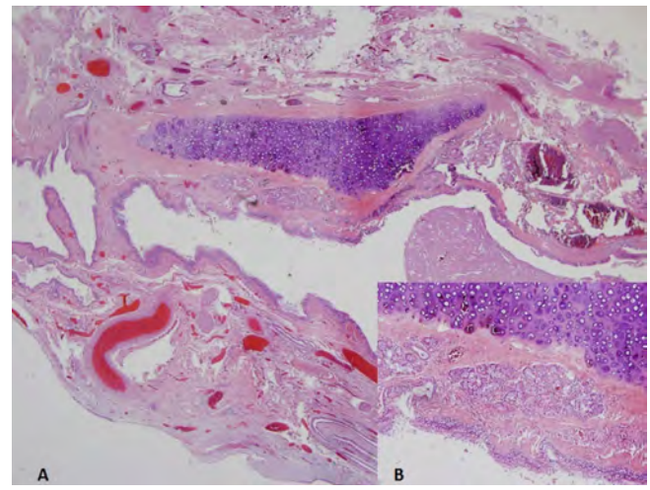


Figure 2. This photomicrograph shows cystic tissue lined by respiratory epithelium (A). There was cartilage and lymphoid tissue in the wall of the cyst (B).

Discussion

Bronchogenic cysts are rare malformations of the ventral foregut due to the abnormal budding of the tracheobronchial tree on the 5th week of fetal development. They are usually reported in the mediastinum and inside the lung parenchyma; rarely, they are reported in extra-thoracic regions including the retroperitoneal region, stomach, pancreas, and gall bladder [1,4,5].

As described by Sumiyoshi et al. in 1985, between the 3rd and 7th weeks of embryonic development, in the presence of delay or insufficiency in pericardioperitoneal canal closure, the elements of the tracheobronchial tree may go through the abdominal cavity. This theory may describe the development of intra-abdominal bronchogenic cysts [1]. These cysts are commonly confused with duplication cysts of the gastrointestinal tract. However, these are lined with ciliated pseudostratified columnar epithelium. Some authors also describe the gastric duplication cysts in the foregut as cystic malformations. Cartilage and glandular tissue is present on the wall of bronchogenic cysts; however, gastric duplication cysts are lined with gastric mucosa [2,4,7]. In both of our cases, the cyst was lined with respiratory epithelium and smooth muscle tissue and sero-mucinous acini were present on the cyst wall. Moreover, in our second case, cartilage tissue was also present on the cyst wall.

Generally, a sub-diaphragmatic bronchogenic cyst is an asymptomatic disease and easily diagnosed. Rarely, it may cause non-specific symptoms such as abdominal pain, nausea-vomiting, and epigastric mass. The exact radiological diagnosis of these cysts is not easy [2-5]. The findings on a CT may resemble gastrointestinal stromal tumors, pancreas cysts, or neurogenic tumors. Retroperitoneal cysts are often confused with lymphangioma, mucinous cyst adenoma, and epidermoid cysts. They are also confused with mucinous carcinoma and retroperitoneal pseudomyxoma due to their cystic and solid components. Endoscopic ultrasound or fine needle aspiration do not give exact results every time. Moreover, aspiration-associated complications such as fistula formation and bleeding have been reported in the literature [5-8]. For that reason, exact diagnosis is made with the histopathological evaluation of resection material, as in our cases.

Surgical excision is recommended in symptomatic cases. Since these cysts have the potential of malignant degeneration, surgery should be considered for a definitive diagnosis. For that reason, in asymptomatic cases, surgery is controversial. However, since preoperative diagnosis is difficult and there are some complications due to percutaneous or endoscopic aspirations, surgery may be an option for both diagnosis and treatment [2,4,7,8].

In conclusion, as in both of our cases, clinical and radiological diagnosis and treatment of extra-thoracic bronchogenic cysts is difficult in the preoperative period. Complete resection is advised in symptomatic cases. In asymptomatic cases, surgery still may be a treatment option for diagnosis, because of possible complications of aspiration and because surgery enables exact histopathological diagnosis. =

Competing interests

The authors declare that they have no competing interests.

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