Hepatic Benign Cystic Mesothelioma in Adult: Case Report of a Rare Hepatic Cyst

Abdullah Mohamed 1, Sherif Elsherif 2, Raafat Makary 3
1. Pathology and Laboratory Medicine, University of Florida College of Medicine – Jacksonville, Jacksonville, USA 2. Radiology, University of Florida College of Medicine – Jacksonville, Jacksonville, USA 3. Neuro-Pathology, University of Florida, Jacksonville, USA

Corresponding author: Abdullah Mohamed, abdullah.mohamed@jax.ufl.edu

Abstract

Benign cystic mesothelioma (BCM), also known as peritoneal inclusion cyst, is a benign mesothelial lined cystic lesion, nearly always described in the pelvis of adult females. Hepatic location of BCM is rarely reported in the literature.

We report a case of hepatic benign cysts in a 65-year-old woman that was incidentally discovered by imaging studies 12 years ago as a small cyst. Recently, the patient started having abdominal discomfort, distension and anxiety. Computed tomography (CT) scan revealed two low-density fluid-filled cystic lesions, the largest in the caudate lobe measuring up to 10.7 cm and causing a mass effect on hepatic veins and inferior vena cava. Laparoscopic marsupialization of the large liver cyst was done without complications. On gross examination, the collapsed cyst wall was thin partly translucent pale tan to pink membranous structure with fine vascularity. No discrete nodularity or solid lesion was or identified. Microscopic examination showed thin fibro-connective wall lined by a single layer of flat cuboidal cells with no cellular atypia. The cyst lining showed characteristic calretinin-positive immunohistochemical reactivity for mesothelium, supporting the diagnosis of benign cystic mesothelioma. Hepatic BCM, is among a broad differential spectrum of cystic liver lesions ranging from developmental, reactive, inflammatory, infectious lesions, benign to premalignant or frankly malignant neoplasms with different treatment strategies. Although BCM is the rarest among the long list of differential diagnosis of hepatic cysts, it’s identification in this rarely reported location is essential to avoid aggressive surgical treatment.

Categories: Pathology
Keywords: cystic lesion, primary liver lesion, peritoneal inclusion cyst, hepatic cyst, benign cystic mesothelioma

Introduction

Benign cystic mesothelioma (BCM, also known as benign inclusion cyst) occurs predominantly in the peritoneum mainly in the pelvis of adult female and very rarely reported in liver. Plaut reported BCM for the first time in 1928, and later, in 1979, Menemeyer and Smit provided a description of the lesion [1]. Despite the unclear pathogenesis, it is considered in some studies as a reactive reaction of the peritoneum to previous intra-abdominal surgeries or inflammation. Other studies suggested benign neoplasm from its tendency to recur and rare malignant transformation [1]. It is usually asymptomatic and is often discovered incidentally during imaging studies performed for other reasons or can cause symptoms such as abdominal pain and distension or mass effect in involved or adjacent organs. We present a rarely reported site for BCM in the liver with clinical presentation, imaging studies and pathological description with a brief literature review.

Case Presentation

A 65-year-old woman presented to the out-patient clinic complaining of abdominal discomfort, fatigue and sleep disturbance. She denied abdominal pain, vomiting or diarrhea. She has a past medical history of invasive ductal carcinoma in the breast and simple small liver cyst. On physical examination, the patient was hemodynamically stable with temperature at 36.8°C, and blood pressure of 147/91 mmHg. Abdominal examination was unremarkable. Abdominal CT scan showed hepatomegaly, steatosis and two low-density fluid-filled intrahepatic cystic lesions, the largest in the caudate lobe up to 10.7 cm causing mass effect compression of hepatic veins and inferior vena cava (Figure 1). Compared to her last CT scan, which was one year ago, the large cyst in the caudate lobe was 7.5 x 6.7 cm. The cysts characteristics in CT favored benign cystic lesion with differential including a simple hepatic cyst and biliary hamartoma.
Abdominal CT scan

Axial arterial phase (a), axial venous phase (b), coronal venous (c), and sagittal venous (d) CT images of the abdomen show large circumscribed hypoattenuating cystic structure (white arrows) within the caudate lobe of the liver measuring up to 10.7 cm causing significant mass effect on the surrounding structures including the IVC (black arrow) and hepatic veins. There is a background of moderate hepatic steatosis. No vascular invasion or lymphadenopathy.

Serum carcinoembryonic antigen was elevated 4.8 (normal < 2.5 in non-smoker) and serum carbonic anhydrase was normal. The elevated serum level of CEA, in the context of the clinical history and CT findings was not raising strong concern for malignancy as it can be non-specific indicator seen in a wide range of lesions, including benign, inflammatory or malignant hepatic and other organ lesions [2].

Due to the symptoms and progressive increase in the cyst’s size, the patient opted for surgical treatment and was admitted for laparoscopic marsupialization. Intraoperative evaluation revealed a non-infiltrative cystic lesion with a delicate plan of cleavage between the cyst wall and hepatic parenchyma. The cyst was aspirated, the fluid sent for culture which came negative, and the wall was completely excised for pathologic examination.

Grossly, the cyst wall was thin partly translucent, pale tan-pink collapsed membranous structure with fine vascularity. No discrete nodules or solid lesions were identified. Microscopic examination showed a thin fibro-connective cyst wall lined by a single layer of flat to low cuboidal cells consistent with benign mesothelium and supported by characteristic calretinin-positive immunohistochemical reactivity [Figure 2]. The lack of cellular atypia or mitoses was consistent with the diagnosis of benign cystic mesothelioma.
FIGURE 2: Microscopic Picture of cyst wall

Collapsed thin cyst wall (A) lined by one layer of flat to low cuboidal cells (B), diffusely reactive for the mesothelial marker (C. Calretinin X5)

The patient’s postoperative period and follow-up evaluations showed complete recovery with no complications and resolution of the clinical symptoms.

Discussion

Hepatic cysts encompass a wide spectrum of lesions including infection from different pathogens (pyogenic, amebic, echinococcal, etc.), pseudocysts, benign, premalignant, primary malignant or metastatic tumors. Pseudocyst may be post-traumatic or from hematoma, seroma or biloma. Developmental cysts include lesions like simple liver cyst, polycystic liver disease, bile duct hamartomas, Caroli disease. Premalignant and malignant cyst lesions include biliary cystadenoma, intraductal papillary neoplasm of bile duct and their malignant counterpart. Imaging studies (US, CT scan and MRI), in most of these lesions display radiological features, which allow a non-invasive presumptive radiographic differential diagnosis. Hepatic BCM is the rarest among the long list of differential diagnosis of hepatic cysts.

Benign cystic mesotheliomas, also known as peritoneal inclusion cyst, are nearly always reported in the pelvis of adult females. The nature of BCM is still controversial between neoplastic versus reactive process. Neoplastic nature is suggested from tendency to recur and rare transformation to malignant mesothelioma [1]. While reactive process was attributed due to frequent association with previous pelvic surgery, pelvic inflammatory disease, or endometriosis [3]. Most of the cases are asymptomatic or have non-specific symptoms. BCM in adult liver is extremely rare, and only one case was found in the English literature [4]. Imaging studies are helpful in limiting the radiographic differential diagnosis to the benign category of cystic lesions. Definitive characterization and cyst behavior are determined from histologic examination of the cyst wall and demonstration of its benign nature along with characteristic positive immunoreactivity staining pattern for mesothelial cell markers as calretinin, cytokeratin, WT1, and/or D2-40. Surgical management is usually not necessary if there is no clinical or imaging concerns or asymptomatic. However, laparoscopic marsupialization or surgical excision may be recommended if the cyst is causing symptoms or complications or a definitive histologic characterization of the cyst is required to exclude underlying infection or malignancy [5,6].

Conclusions

Benign cystic mesothelioma is extremely rare in the liver with non-specific clinical presentation. Definitive cyst characterization and behavior are determined by histologic examination for demonstration of its benign mesothelial nature. Although BCM is the rarest among the long list of differential diagnosis of hepatic cysts, it’s identification in this rarely reported location is essential to avoid aggressive surgical treatment.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Abdullah Mohamed

Acquisition, analysis, or interpretation of data: Abdullah Mohamed, Sherif Elsherif, Raafat Makary

Drafting of the manuscript: Abdullah Mohamed, Sherif Elsherif

Critical review of the manuscript for important intellectual content: Abdullah Mohamed, Raafat Makary
Supervision: Raafat Makary

Disclosures

Human subjects: All authors have confirmed that this study did not involve human participants or tissue.

Conflicts of interest: In compliance with the ICMJE uniform disclosure form, all authors declare the following: Payment/services info: All authors have declared that no financial support was received from any organization for the submitted work. Financial relationships: All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. Other relationships: All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

References


