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Differential Diagnosis Difficulty between Biliary Cystadenoma and Cystaadenocarcinoma of the Liver: A Case Report

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Abstract

Biliary cystadenoma is a multilocular cystic benign tumor which arises in the intrahepatic biliary system. Diagnosis is often difficult and based on imaging, however histology remains the only argument for diagnosis certainty. We report a case of biliary cystadenoma in a 69-year-old female. Observation: it was a 69-year-old female with history of RHUPUS syndrome (Lupus + Rheumatoid arthritis) under low-dose corticosteroid therapy. She presented with pain in the right hypochondrium with permanent moderate intensity for 3 months associated to altered performance status. Physical examination showed a hepatomegaly, cervical subluxation, and pulmonary condensation syndrome. Hepatobiliary ultrasound showed presence of multiple septate cystic formations occupying segments 1, 7, 8, 5 of the left liver. Abdominal CT scan and hepatic MRI were in favor of biliary cystadenoma but could not rule out biliary cystadenocarcinoma with certainty. Conclusion: biliary cystadenoma is a benign cystic tumor of the liver. The differential diagnosis with cystadenocarcinoma is most often difficult and usually requires histological analysis for diagnostic certainty.

Keywords

Cyasdenoma, Cysadenocarcinoma, Bile Duct

1. Introduction

Biliary cystadenoma is a rare, benign, cystic and multilocular tumor which arises the intrahepatic biliary system occurring most often in middle-aged women [1]. Diagnosis is often difficult due to a lack of clinical, biological, and radiological

specificity. Imaging remains the key element of orientation, however confirmation remains histological.

We report a case of intrahepatic biliary cystadenoma in a 69-year-old female based on imaging after ruling out other causes of liver benign cystic tumors. However, the problem of differential diagnosis with a biliary cystadenoma carcinoma arose due to the lack of histological evidence.

2. Observation

It was a 69-year-old female, Gravida 10, Para 8, 2 abortions, with history of RHUPUS (Lupus and rheumatoid arthritis) with follow-up for a year in the internal medicine department of the university hospital Aristide Le Dantec, under low corticosteroid therapy dose (prednisone 5mg/day) and under hydroxychloroquine (400 mg/day). She presented for 3 months with pain in the right hypochondrium with permanent moderate intensity aggravated by coughing and deep inspiration and associated with bowel movement disorder as of constipation, and productive cough with white phlegm. On admission, physical examination found a renitent painful hepatomegaly with an irregular anterior surface, a blunt lower edge, cervical subluxation, syndromic anemia, and a grade 2 altered performance status. Biology tests found a non-specific inflammatory response syndrome with elevated CRP level of 48 mg/l and inflammatory anemia with hemoglobin level of 9.5 g/dl.

Liver assessment was normal apart from the slightly elevated GGT at 2 N, the alpha fetoprotein was normal at 1.15 ng/ml.

Hepatobiliary ultrasound showed a hepatomegaly with regular contours, heterogeneous echostructure by the presence of multiple compartmentalized cystic formations occupying segments 1, 7, 8, 5, the largest of which sat at the level of the left liver 89 × 68 mm and exerted a mass effect on the left branch of the portal vein and pancreas (**Figure 1**). There was no ascites, and the bile ducts were not dilated.

Confronted to these ultrasound signs and the endemic context, the hypothesis of a hydatid cyst had been brought up but was unlikely, as the analysis of the cyst

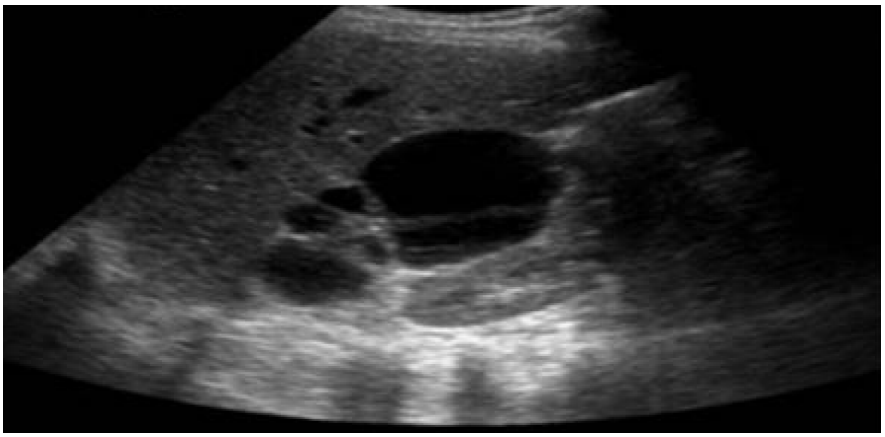

Figure 1. Ultrasound of septate cystic lesions of the liver.

fluid obtained through ultrasound-guided puncture, under antiparasitic cover with albendazole did not highlight the presence of laminar membrane debris, hooks or protoscolex after cytological examination. The hydatid serology by immunoelectrophoresis was negative.

Amoebic and pyogenic abscess, as well, were eliminated after bacteriological and parasitic study of the liquid did not isolate any germ. The amoebic serology was negative. Abdominal CT showed multiple formations of fluid density disseminated over the entire hepatic parenchyma, the largest of which measured 119 × 52 mm. Some were compartmentalized and not enhanced by contrast and moderately compressing the hepatic hilum, suggesting a biliary cystadenoma (**Figure 2**).

Hepatic magnetic resonance imaging showed well-limited diffuse septate cystic formations located in the left liver with hyposignal on T1 and homogeneous hypersignal on T2, with non-enhanced septa after injection of gadolinium, suggesting a biliary cystadenoma.

The diagnosis of biliary cystadenoma was retained; however, liver biopsy would have enabled to eliminate with certainty a cystadenocarcinoma. The patient was put under analgesic treatment, a left hemi-hepatectomy was indicated, but the patient died of a pulmonary embolism that occurred on day 12 of hospitalization.

3. Discussion

Biliary cystadenoma is described as a multilocular cystic benign tumor with smooth surface and external vascularization. It has recurrence characteristic and potential evolution towards malignancy [1]. The tumor arises in the biliary system most often from the intrahepatic bile ducts and very rarely from the extrahepatic bile ducts or the gallbladder.

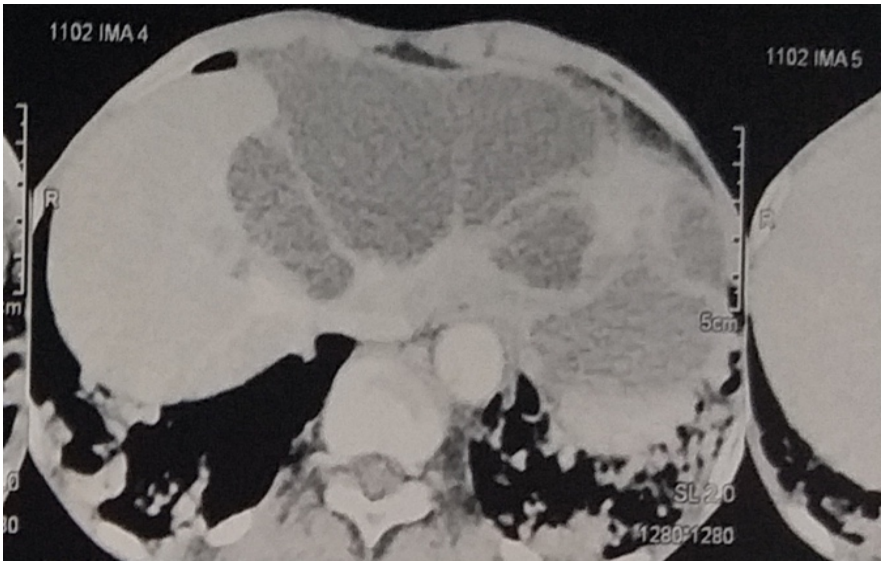

Figure 2. Septate hypodense lesions on CT scan.

The origin of the cystadenoma is often disputed but for most, it would be of congenital origin originating from the primitive ectopic biliary tissue [1] [2]. It is a rare tumor and represents 5% of benign liver tumors and occurs in 80% of cases in middle-aged women over 50 - 60 years [3] [4].

The clinical signs are not very specific, there may be abdominal pain in the right hypochondrium or in the epigastric area of varying intensity, sometimes bearable, jaundice in the eventuality of compression of the bile ducts, ascites secondary to portal hypertension resulting from compression of the portal vein [3]. In our case, the revealing signs were pain in the right hypochondrium and hepatomegaly.

The onset of ascites, jaundice or exacerbation of pain should raise concern over complications such as intracystic hemorrhage, secondary infection, peritoneal rupture, or malignant transformation. On ultrasound, the typical lesion is a large, sometimes unilocular, multilocular cyst and may be anechoic, hypoechoic, or echogenic and septate [3] [4]. The abdominal CT scan provides further information on the nature of the cystic lesion and enables locoregional assessment by specifying the site of the tumor, its size and its relationship with the neighboring organs. The lesions are often hypodense with a fluid density (less than 30 Hounsfield) with rare septal and parietal calcifications [5] [6]. After the injection of contrast product, a check shall will be made of an enhancement of the capsule, of the septa, the presence or not of nodule, the local invasion of the liver, the bile ducts or the portal vessels orienting towards a cystadenocarcinoma [6].

In our patient, the abdominal CT did not show any calcifications nor septa nor nodules and the wall of the cysts was regular and thin. Magnetic resonance imaging is more sensitive than CT scan, showing a hypointense lesion on T1 and strongly hyperintense and heterogeneous on T2 [7]. In our patient, the cysts were homogeneous on T2 on MRI. In pathological anatomy findings, tumors appear bulky with an average diameter varying between 7 and 15 mm, multilocular, separated by partitions and surrounded by a fibrous capsule. The contents are liquid with a viscous, whitish, yellowish, bilious, or greenish. Hemorrhagic contents should suggest cystadenocarcinoma [7] [8]. Histological examination shows lesions with mucinous differentiation with or without mesenchymal stroma [7]. In our case, the liquid was viscous and opalescent, histological examination did not find cells suspicious for malignancy.

Treatment remains straightaway radical due to the risk of progression to malignancy and the risk of recurrence. The lesion must be removed while respecting a margin of healthy tissue because of the impossibility of differentiating, on a macroscopic level, cystadenoma from a cystadenocarcinoma, hence the strict observance of oncological measures during surgery [4] [9].

4. Conclusion

Biliary cystadenoma is a benign cystic tumor of the liver, its clinical and radiological semiology is nonspecific even if the imagery directs, the diagnosis of cer-

tainty remains histological. Differential diagnosis with cystadenocarcinoma is often difficult, hence the importance of a histological examination for diagnostic certainty.

Provenance and Peer Review

All authors have read and approved the document.

Consent

Patient gave consent to report case.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

References

[1] Ammar, M. and Noomen, F. (2015) Cystadénome hépatique simulant un kyste hydatique. *The Pan African Medical Journal*, **20**, Article No. 294. https://doi.org/10.11604/pamj.2015.20.294.6644

[2] Brittingham, C. and Tuma, F. (2020) Treasure Island. StatPearls Publishing.

[3] Alahyane, A., El Fahssi, M., Elhjouji, A., *et al.* (2010) Hepatic Cystadenoma: The Need for Radical Treatment. *Journal Africain du Cancer*, **2**, 272-274. https://doi.org/10.1007/s12558-010-0118-9

[4] Bassou, D., Darbi, A., Goasdoué, P. and El Kharras, A. (2007) Intrahepatic Biliary Cystadenoma: Contribution of Imaging. *Leaflets of Radiology*, **47**, 387-391.

[5] Filippi De La Palavesa, M.M., Wong, C., Vasilescu, C., *et al.* (1998) Imaging of Cystic Tumors of the Liver. *Leaflets of Radiology*, **38**, 162-174.

[6] Boytchev, I., Georgelin, M., Bedossa, P. and Buffet, C. (1999) Intrahepatic Biliary Cystadenocarcinoma. *Gastroentérologie Clinique et Biologique*, **23**, 981-983.

[7] Edrani-Canihac, M., Le Bail, B., Rivel, J., *et al.* (2000) Cystadenoma Hepatobiliary with Mesenchymal Stroma a Hormone—Dependent Tumor? *Annals of Diagnostic Pathology*, **20**, 14-18.

[8] Soares, K.C., Arnaoutakis, D.J., Kamel, I., Anders, R., Adams, R.B., Bauer, T.W., *et al.* (2014) Cystic Neoplasms of the Liver: Biliary Cystadenoma and Cystadenocarcinoma. *Journal of the American College of Surgeons*, **218**, 119-128. https://doi.org/10.1016/j.jamcollsurg.2013.08.014

[9] Lewis, W.D., Jenkins, R.L., Rossi, R.L., Munson, L., ReMine, S.G., Cady, B., *et al.* (1988) Surgical Treatment of Biliary Cystadenoma. A Report of 15 Cases. *The Archives of Surgery*, **123**, 563-568.