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Hemorrhagic Hepatic Cyst mimicking a Biliary Neoplasm in a patient with Polycystic Liver Disease: Report of a Case with emphasis on Contrast-Enhanced Ultrasound with SonoVue®.

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Written informed consent was obtained from patient according the Ethical Committee

of our Institution.

TYPE OF MANUSCRIPT: Case report.

Abstract

The occurrence of intracystic hemorrhage in benign liver cysts is usually seen in huge solitary

cysts and in older individuals. We report a case of multiple simple hepatic cysts with

intracystic hemorrhage complicating one of the cysts in whom the correct diagnosis was

accurately made by Contrast-Enhanced Low-MI Real-Time Ultrasound (CEUS) Imaging with

Sonovue® (Bracco, Milano, Italy). Our report emphasize on the crucial role of CEUS as

imaging modality in providing the informations needed to differentiate haemorragic cysts

from cystic liver tumors such as cystadenoma and cystadenocarcinoma, potentially avoiding

the use of more invasive and expensive imaging modalities such as CT or MRI imaging.

Keywords: Contrast-enhanced Ultrasound (CEUS); Haemorragic cysts; Polycystic liver

disease; Complex Cystic Liver Lesions; Microbubbles;

Introduction

Simple liver cysts are uncommonly complicated by hemorrhage, which is usually seen in

huge solitary cysts, frequently in older individuals. Therefore, intracystic hemorrhage from

one of multiple cysts has rarely been reported worldwide [1, 2].

Differential diagnosis of a hepatic cyst with intracystic hemorrhage from a hepatobiliary

cystic tumour is very difficult exclusively on the basis of clinical and US features causing a

diagnostic dilemma if one is not aware of the potential diagnoses and imaging pitfalls [3].

Real-time contrast-enhanced ultrasound (CEUS) using second-generation ultrasound contrast

agents (UCAs) is a "new" simple, immediate, and effective tool in investigating FLLs [4, 5].

In our institution, US is the initial imaging modality of choice for the evaluation of liver, and

CEUS with SonoVue® is usually chosen to characterize FLLs indeterminate on conventional

US [6].

We herein present a patient with spontaneous intracystic bleeding of a large simple hepatic

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cyst that mimicked a cystic biliary tumour, correctly diagnosed by Contrast-Enhanced Low Mechanical Index (MI) Real-Time Ultrasound (CEUS) Imaging with Sonovue® (Bracco, Milano, Italy).

Case Report

A 34-year-old woman with polycystic liver disease had been under follow-up with laboratory examination and ultrasound (US) for two years. She was admitted to our institution for the complaint of vague upper abdominal pain and abdominal discomfort. Clinical examination was unremarkable. Laboratory tests and tumor markers were also normal, except for a mildly abnormal liver function.

Abdominal US showed a complex cystic lesion with slighly irregular walls and hyperechoic intracystic structures measuring 12 x 9 cm in VIII hepatic segment. In addiction, other simple cysts involving both liver lobes were also sonographically detected.

To better assess the complex cyst, the patient was submitted to Contrast-Enhanced Low-MI Real-Time Ultrasound (CEUS). The agent employed was Sonovue® (Bracco, Milan, Italy), a sulfur hexafluoride-filled microbubble contrast medium. It was injected into the antecubital vein in bolus bolus fashion of 4.8 mL followed by a flush of 10 mL of 0.9% normal saline solution. The single bolus was split into two injections, one for each liver lobe. We waited at least 5 min between the two injections. For both injections we scanned the largest and/or the most suspicious lesion during the arterial phase and then explored the remaining lesions within the same lobe for up to 5 min. We employed a dual-frame real-time mode with the fundamental mode US image on the right part of the monitor and the contrast-enhanced mode image on the left. The ultrasound beam was focused at the deeper aspect of the lesion examined. After contrast injection, continuous scanning began immediately and lasted 4-5 min.

CEUS imaging confirmed the diagnosis of hemorrhagic cyst because the intracystic structures solid-appearing at baseline US were showed to be avascular throughout all phases (**Fig. 1**), suggesting blood clots and fibrin strandings due to intracystic hemorrhage.

Because of the high-risk of the enlargement and rupture of the haemorragic cyst, surgery was considered to be indicated. Therefore, a contrast-enhanced Computed Tomography (CT) was performed for a preoperative assessment.

On CT, the lesion appeared as a homogeneously low-density lesion. However, the intracystic structures detected on US were not visualized after intravenous administration of contrast medium. Finally, the presence of multiple other simple cysts was confirmed (**Fig. 2**).

At surgery, a total removal of the lesion was easily performed. As expected, the resected lesion was histopathologically proved to be a benign liver cyst containing multiple blood clots and fibrin strandings. Histopathology showed fibrocollagenous cyst wall lined by one row of cuboidal epithelial cells suggestive of a simple cyst.

The patient had an uneventful recovery and was discharged 14 days after the surgery.

Discussion

Simple cysts are easily diagnosed with US as well-defined anechoic lesions with posterior acoustic enhancement and imperceptible walls [7, 8]. However, when a liver cyst contains a large amount of blood clot and fibrin deposits, which is a not common condition, the lesions may exhibit internal echoes that mimic septations or solid portions during US imaging, resulting in difficulty in differentiating such lesions from cystic liver tumors such as cystadenoma and cystadenocarcinoma. Additionally, it should be noted that intracystic bleeding can occur also in cystic liver tumor [9].

Thus, since the clinical implications and therapeutic strategies vary considerably, the radiologist have to know such US features in order to indicate a correct imaging work-up [10].

US is the first choice technique, in liver imaging, in view of its safety, widespread availability and low cost [11, 12]. Unfortunately, the reliability of conventional US in characterizing complex cystic focal liver lesion is limited and diagnosis is especially challenging [13], thus, there has been for years a tendency for clinicians to prefer contrast-enhanced computed tomography (CECT) or magnetic resonance imaging (MRI) in evaluating these lesions.

To date, the newly developed low-MI real-time CEUS techniques using the second generation of UCA has led to an accurate depiction of either macro- or microvasculature, thus offering information about contrast enhancement of liver and FLLs, almost likewise CT and MRI [11, 12]. As expected, CEUS has proven to be extremely useful in the evaluation of both solid liver lesions and complex renal cysts [14, 15]. CEUS is also highly accurate in the categorization of complex cystic FLLs as benign or malignant, with a sensitivity and specificity ranging, respectively, from 87% to 93% and from 88% to 98% depending on the

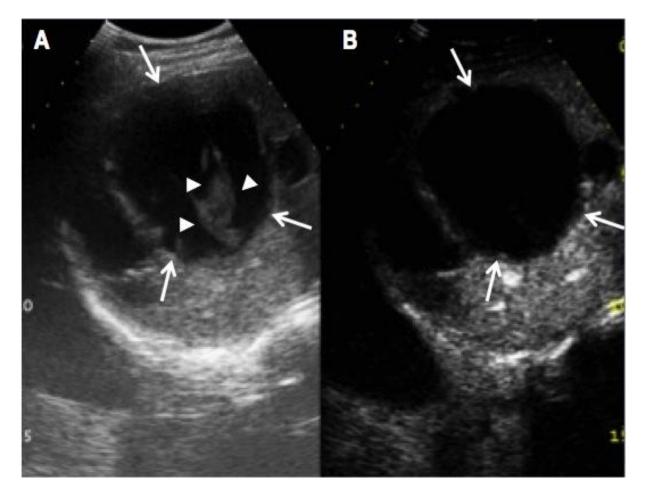
operator's experience [13]. However, to our knowledge, little experience exists about CEUS imaging with Sonovue® in differentiation of biliary cystic neoplasms from haemorragic hepatic cysts [16, 17].

CEUS can contribute to a reliable differential diagnosis, thus ruling out neoplasms. Indeed, when are destroyed by ultrasound, microbubbles of US contrast agent enable to visualize vascular flow within septa or solid components of complex cysts, which is absent in simple cysts with intracystic haemorrhage [6, 18].

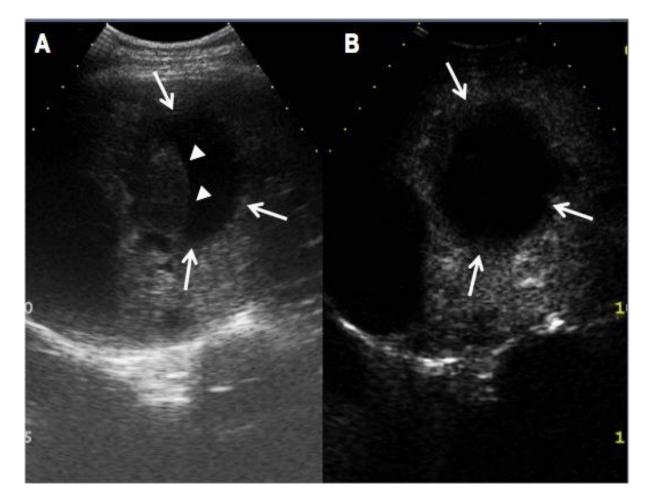
Even though cystic lesions at US are uncommon and the differential diagnosis is not easy, the solid nature should be always suspected. Therefore, we have developed an evidence-based diagnostic algorithm to guide clinicians and radiologists in correctly characterizing these lesions (**Fig.3**).

In conclusion, although most simple hepatic cysts are asymptomatic, complications of hepatic cysts may occur and differential diagnosis from cystic biliary tumour should always be considered. In this setting, CEUS imaging should be performed when a diagnosis of haemorragic cyst is suspected, potentially avoiding the use of more invasive and expensive imaging modalities such as CT/MR, which often may be not diagnostic. This is an added value in countries where US is regarded as the first choice modality for liver survey. The correct diagnosis leads to the best management for the patient.

FIGURE



1a



1b

Figure 1 (**a**, **b**). Split-screen mode with fundamental US image on the left side of monitor and CEUS image on the right. Conventional US showing a complex cystic lesion (arrows) containing hyperechoic intracystic structures (arrowheads). Serial CEUS images obtained at 70s (**a**) and 108s (**b**) after UCA injection show the homogeneous nonenhancement of the lesion (arrows) confirming the diagnosis of hemorrhagic cyst.

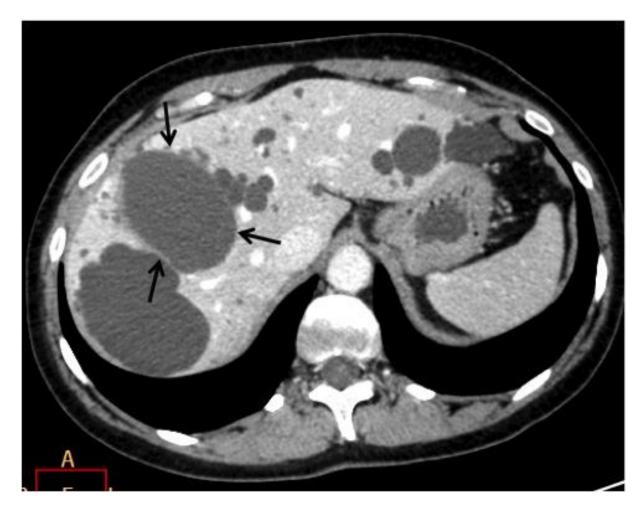


Figure 2. Contrast-enhanced CT in portal phase showing the haemorragic cyst as a homogeneously low-density lesion. The intracystic structures detected on US were not visualized after intravenous administration of contrast medium (arrows). Multiple hypoattenuating simple cysts without enhancement were also detected in both liver lobes.

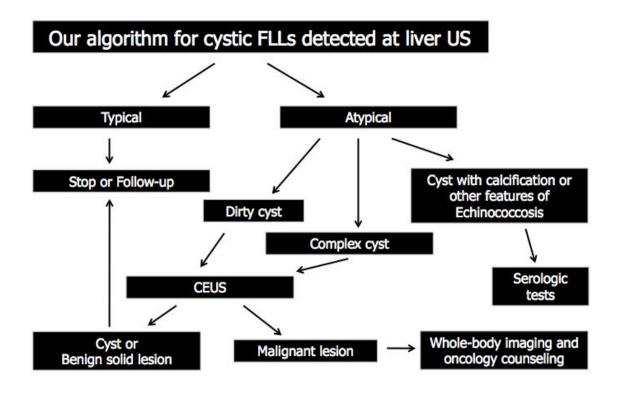


Figure 3. Flow-chart for cystic FLLs detected at US that we have adopted in our institution.

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