



Full Length Research Article

BILIARY HAMARTOMA MIMICKING PNEUMOBILIA: A CASE REPORT

***Hedfi Mohamed, Laabidi Asma, Abdelaali Imen, Ikram Messaoudi, Dougui Hedi and Chouchene Adnene**

Surgery Department, Gastro-Enterology Department, FSI Hospital Tunisia

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ABSTRACT

Multiple biliary hamartomas (MBH) are a rare cause of multiple benign hepatic lesions. The condition is also known as von Meyenburg complexes, multiple bile duct hamartomas and biliary microhamartomas. MBH is asymptomatic and usually found incidentally, where it is important to differentiate from other causes of multiple liver lesions, particularly metastases. In this report, a case of biliary hamartoma mimicking pneumobilia is presented.

INTRODUCTION

Biliary hamartoma is a rare benign hepatic tumor. It is also known as von Meyenburg complex. Pathologically, it consists in cystic dilated bile duct embedded in a fibrous stroma (Wei et al., 1997). Radiology, such as ultrasonography (US) and computed tomography (CT) could help suspect diagnosis. Although careful analysis of magnetic resonance images could permit diagnosis of von Meyenburg complexes, referral for open biopsy to exclude liver metastases is required in suspicious cases. In this report, a case of biliary hamartoma mimicking pneumobilia is presented.

Case report

A 55-year old man with no significant past medical history presented in our clinics with the complaint of right hypochondrium band-like pain lasting over the preceding 3 months. Physical examination revealed no specific findings such as hepatomegaly. Liver tests were within the normal range: the serum level of alanine aminotransferase (ALT) was 25 IU/L, aspartate aminotransferase (AST) was 16 IU/L and alkaline phosphatase (ALP) was 142IU/L. Total bilirubin was 8 mg/L and gamma-glutamyl-transpeptidase was 34IU/L.

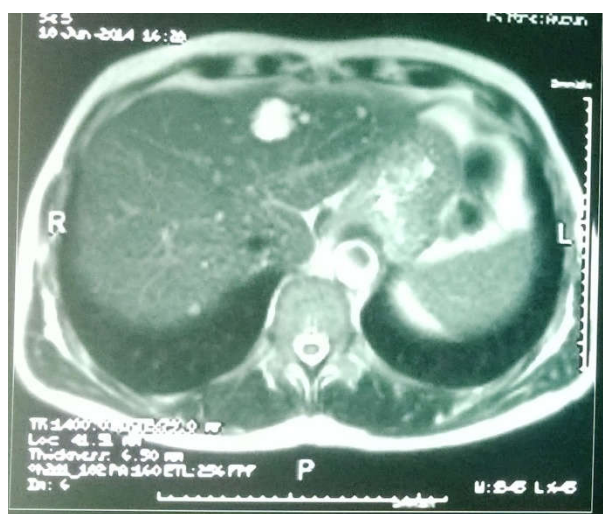
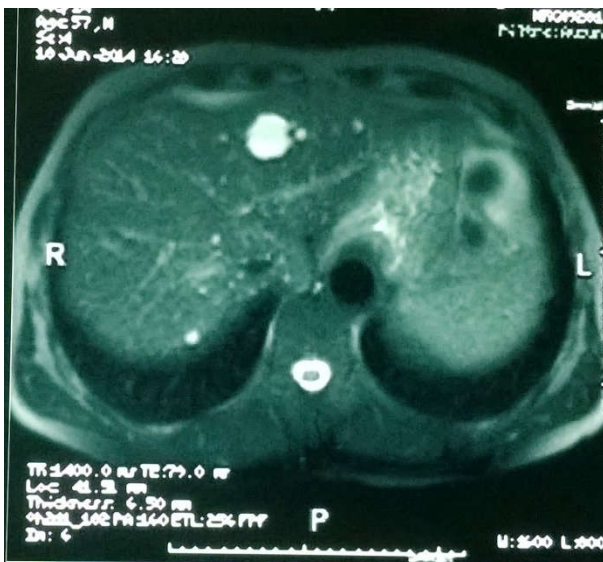
Prothrombine rate was 100%. Alfa-foetoprotein was not evaluated. Ultrasound of the liver was performed and revealed small cystic dilations of intrahepatic bile ducts in the liver segments III and VI that was compatible with the diagnosis of biliary cysts. It also showed highly reflective linear echoes with multiple discrete bubbles in the duct and faint shadowing. These bubbles were mobile on real time. Pneumobilia resulting from a biliary-digestive fistula was suspected. For further evaluation, magnetic resonance examination was performed. It confirmed the presence of biliary cysts and revealed tiny nodules with different size that were scattered in the liver. These hepatic nodules showed low signal intensity on T1-weighted image, and high signal intensities on T2-weighted image and diffusion image. After contrast enhancement with Gadolinium, subtle rim-like enhancement was noticed. No communication with the intrahepatic bile ducts was shown. Diagnosis of Von Meyenburg complexes was made (photos 1,2,3). Needle biopsy of the liver was feared since some biliary cysts were arranged along the needle path.

DISCUSSION

Biliary hamartoma are uncommon benign tumors of the liver and are thought to reside on the spectrum of congenital hepatic fibrosis.

***Corresponding author: Hedfi Mohamed,**

Surgery Department, Gastro-Enterology Department, FSI Hospital Tunisia.



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The pathogenesis of these lesions is the result of arrested development and remodeling of the small, peripheral intrahepatic biliary ducts (Principe *et al.*, 1997).

Biliary hamartoma is usually multiple and small well circumscribed nodules ranging from 1 mm to 1.5 cm. However, in some cases, it may present as a single mass (Yong Moon Shin, 2011). Although the risk of malignant conversion is low, malignant transformation into cholangiocarcinoma has been reported (Orii *et al.*, 2003). Affected patients are usually asymptomatic and have no laboratory abnormalities. Biliary hamartomata may appear hypoechoic, hyperechoic, or as a mixed echotexture on ultrasonography and the nodules can accompany comet tail artifact (Zheng *et al.*, 2005). On contrast enhanced CT, the lesions look like hypodense nodules of variable enhancement and may demonstrate an irregular border (Lung and Phillip, 2013).

Based on CT scanning alone, VMC cannot be definitively differentiated from metastatic disease. Then MRI is often performed to differentiate biliary hamartomas from liver metastases. Typically, VMCs appear hypointense on T1-weighted imaging and are hyperintense on heavily T2-weighted sequences with similar signal intensity to the spleen, but less than liver cysts (Mortele *et al.*, 2002). In the case we reported, right hypochondrium band-like pain was the revealing symptom of biliary hamartomas which is unusual. Ultrasonography did not help suspect diagnosis as comet tail artifact, a common US sign of Von Meyenburg complexes, was interpreted as pneumobilia. Then, MRI was performed to confirm pneumobilia. However, MRI showed no pneumobilia but typical features of VMCs. Diagnosis was made on the basis of MRI findings. So, most of biliary hamartomatosis does not require pathological confirmation nowadays when it presents typical radiological appearance. However, when the lesion is suspicious, tissue confirmation is mandatory.

Conclusion

This report highlights the performance of MRI as diagnostic tool of biliary hamartoma. In fact for many years, diagnosis of biliary hamartoma has been frequently not considered until after liver biopsy. Although, there is no guidelines for the management of biliary hamartoma yet, no treatment is required in most cases. However, as the risk of malignant conversion exists, regular imaging monitoring of the hepatic lesions should be performed.

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