



Incidental Discovery of von Meyenburg Complexes: A Case Report on Biliary Microhamartoma

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Abstract

Biliary hamartoma or von Meyenburg complex (VMCs) is a rare benign congenital malformation of the biliary duct [1]. It is often seen incidentally on imagery or surgery as multiple small subcapsular nodules scattered throughout the liver [2] or uncovered by autopsy. Detecting by imaging modalities is thought to be uneasy because of their asymptomatic nature and small size [3].

Although VMCs are rare, they are easily confused with metastatic diseases of the liver on imaging [4]. Biliary hamartomas do pose a minor risk of malignant transition to intrahepatic cholangiocarcinoma or, less commonly, hepatocellular carcinomas [5]. Because of its rarity and diverse clinical presentation, management of this condition can be challenging [6,7].

Keywords: Hepatocellular carcinomas; Biliary microhamartoma; Biliary duct; Autopsy

Introduction

Biliary microhamartoma also called von Meyenburg Complexes (VMCs) is a rare condition discovered accidentally in asymptomatic patients. This condition was first described by von Meyenburg in 1918. Biliary microhamartomas incidence ranges from 0.5% to 5.6% in autopsy studies and the size of the lesions ranges from 0.1 to 1.5 cm in diameter [8].

VMCs are caused by ductal plate malformations of the smallest intrahepatic bile ducts. VMCs have been associated clinically with a variety of symptoms, therefore posing diagnostic challenges, especially at the time of their initial presentation. However, symptomatic lesions are uncommon and giant lesions are exceedingly rare. When encountered, they should be excised because there are reports of malignant changes in large, symptomatic lesions. The presence of multiple liver cystic lesions can occasionally represent a diagnostic dilemma. Multiple simple liver cysts are by far the most common etiology. Occasionally, however, multiple parasitic liver cysts or Caroli's disease can cause diagnostic uncertainties; rarely, liver metastatic disease, causing necrosis of the affected liver parenchyma, must be excluded.

There is an association between biliary microhamartoma and hepatic malignancies, especially the intrahepatic cholangiocarcinoma [8]. In addition, the association between it and the adult type of polycystic liver disease was also reported. However, there is no correlation between the number of biliary microhamartoma and the liver age or weight [9]. Biliary microhamartoma is usually diagnosed by imaging studies as MRI. However, some cases are

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confused with the metastatic disease of the liver on US or CT, so the gold standard for diagnosis in such cases is the liver biopsy [10].

Case Presentation

A 55-year-old male with a medical history of hypertension and type 2 diabetes mellitus presented to the outpatient clinic with intermittent, non-specific right upper quadrant abdominal pain. This discomfort had been ongoing for the past three months. He denied any associated symptoms such as fever, jaundice, weight loss, or changes in bowel habits. On physical examination, the patient appeared well and in no acute distress. His vital signs were within normal limits: blood pressure 130/85 mmHg, heart rate 72 bpm, respiratory

rate 16 breaths/min, and temperature 36.8°C.

Abdominal examination revealed mild tenderness in the right upper quadrant but no palpable masses or hepatomegaly. There was no evidence of ascites or peripheral edema. Laboratory investigations showed normal results: complete blood count (CBC), liver function tests (LFTs) including alanine transaminase (ALT) 35 U/L, aspartate transaminase (AST) 30 U/L, alkaline phosphatase (ALP) 90 U/L, and total bilirubin 0.8 mg/dL. Serum albumin was 4.2 g/dL and renal function tests were within normal limits (Figure 1). Abdominal ultrasound revealed multiple small hyperechoic lesions scattered throughout the liver, raising the possibility of metastatic disease. No evidence of biliary obstruction or gallstones was noted.

Cross Sectional Imaging findings of biliary microhamartomas - von Meyenburg Complex

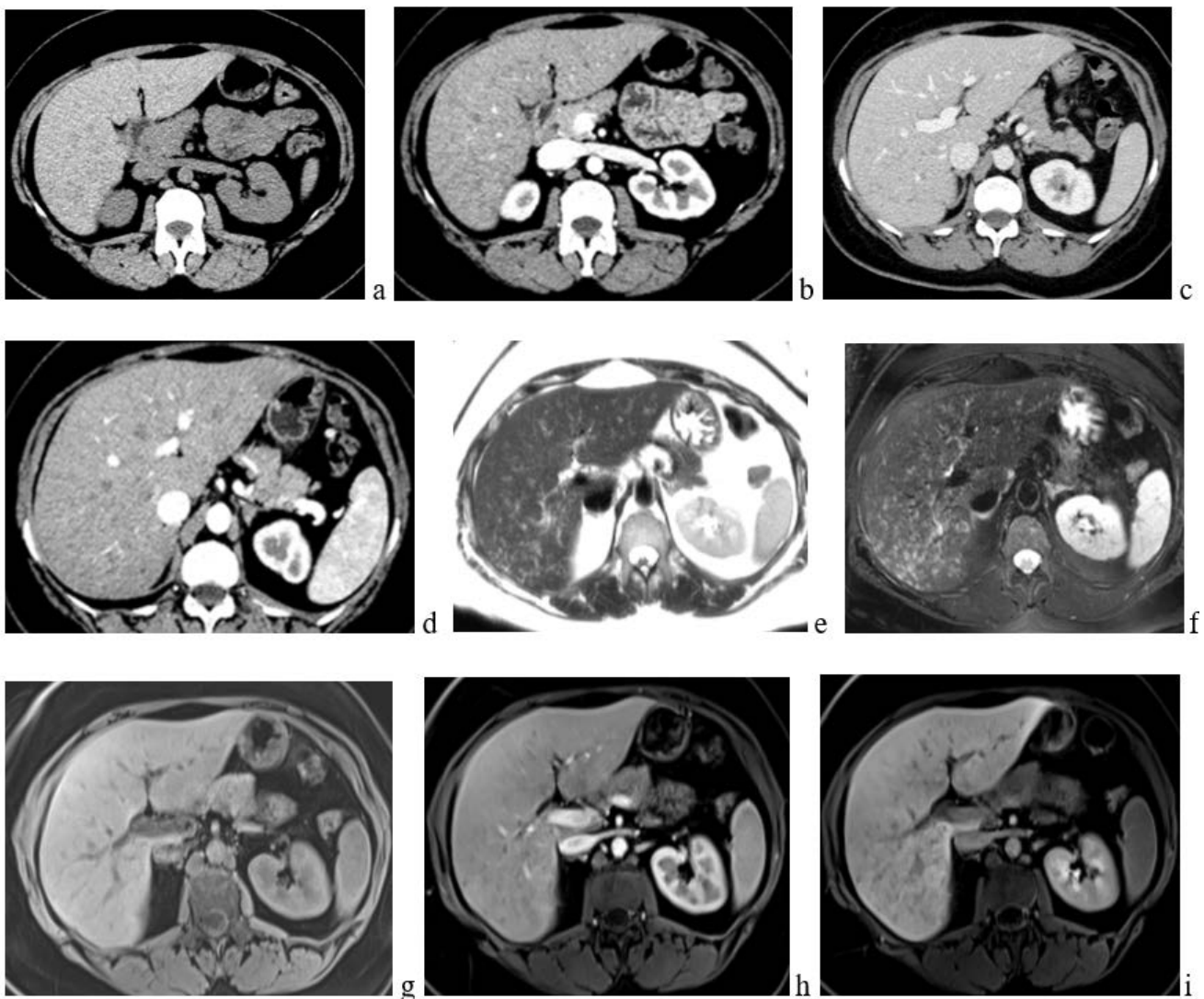


Figure 1: **a:** There are multiple well-defined lesions scattered throughout the liver, smaller than 10 mm each. The non contrast and **b,c,d:** contrast-enhanced CT scan through liver shows small hypodense lesions of uniform size and no enhancement after i.v. iodine contrast administration. **ef:** Most lesions are in right lobe. The lesions in the non-contrast and contrast-enhanced MRI through liver show high signal intensity on T2-WI, **g:** low signal intensity on T1-WI, and **h,i:** no enhancement after gadolinium administration.

Discussion

VMCs are thought to be caused by embryologic abnormalities of the ductal plates which are responsible for the development of most peripheral intrahepatic ducts [11]. Macroscopic hepatic cysts, Caroli disease, congenital hepatic fibrosis, and polycystic kidney disease were found to be correlated with VMCs [11,12]. Generally, these formations comprise bile duct remnants that incorporate small biliary ducts inside a fibrous stroma, often containing thickened bile.

A study showed that a total of 6 (0.35%) out of 1697 liver biopsies were found to have multiple biliary hamartomas [13]. While 5.6% of multiple biliary hamartomas were found in an autopsy study of 2843 cases [12]. Interestingly, VMCs were frequently seen in older patients and could be an acquired lesion that is associated with liver cirrhosis [12,14]. However, it is still unclear why VMCs are also seen in noncirrhotic patients as well [13].

VMCs are usually diagnosed in imaging studies when there are multiple small (usually <10mm) cysts scattered throughout the liver as patients with VMCs are often asymptomatic and have normal liver function tests. While some patients may not survive, such as those with autosomal recessive polycystic kidney disease who typically do not survive beyond early infancy, others may be asymptomatic and discovered incidentally during laparotomy or autopsy [15]. They could also present non-specific abdominal symptoms. Rarely, patients can present with infectious complications [16]. VMCs are generally considered incidental findings and could mimic hepatic metastases. The imaging characteristics span a broad spectrum, ranging from solid to a combination of solid and cystic, to purely cystic lesions. Typical imaging features that are often indicative of VMCs include the presence of multiple small comet-tail echoes on ultrasound, scattered hypodense lesions throughout the liver that do not enhance on computer tomography imaging, and cystic structure with normal extrahepatic and intrahepatic bile duct on magnetic resonance imaging (MRI) and magnetic resonance cholangiopancreatography (MRCP) [10,17]. The presence of typical imaging findings is sufficient to make a correct diagnosis even without requiring histological confirmation. MRI and MRCP are helpful to differentiate VMCs from other diseases. Due to the nonspecific imaging appearance, the differential diagnosis of VMCs is broad. Ruling out of malignancy or metastases is crucial. If the imaging results raise suspicion, a biopsy may be required.

VMCs have been shown to be associated with malignant disease, especially cholangiocarcinoma [5,18,19]. A study showed that the involvement of APC, p53, p19, and PTEN oncogenes correlates with an early event in the development of cholangiocarcinoma in patients with VMCs [18]. However, given the restricted sample size analyzed in the study, it is difficult to detect any specific mutation. Interestingly, through

an animal model of aflatoxin-induced cholangiocarcinoma, it was observed that the progression of tumors involved an intermediate phase of cystic biliary proliferation that closely resembles VMCs [20]. This finding further strengthens the idea that VMCs could serve as the precursors for sporadic cholangiocarcinoma. However, a more detailed analysis with a larger number of cases is required to advance our understanding of cancer progression in cholangiocarcinoma.

To date, there is no established follow-up protocol for patients with multiple VMCs due to their rarity. When multiple VMCs are present, it is highly recommended to carefully examine for the possibility of coexisting cholangiocarcinoma. Screening for cholangiocarcinoma in patients with VMCs could be done as well as this population has a high risk of developing cholangiocarcinoma.

Conclusion

Biliary microhamartoma, also called von Meyenburg complexes (VMCs), presents a rare congenital biliary duct malformation. Though mostly asymptomatic, VMCs can mimic metastatic liver disease on imaging, often requiring liver biopsy for histological confirmation of uncertain cases. Their link to hepatic malignancies, particularly intrahepatic cholangiocarcinoma, highlights the need for vigilant monitoring, especially when multiple lesions are present. Further research is necessary to understand the exact mechanisms underlying the progression of VMCs to malignant tumors and to establish follow-up protocols. Screening for cholangiocarcinoma in VMC patients is advised due to increased malignancy risk in this population. Despite its rarity, clinician awareness of VMCs is essential for prompt diagnosis and management, ultimately improving patient outcomes.

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