

中文題目：罕見的肝臟多結節性疾病：膽道過誤瘤—病例報告

英文題目：An unusual polynodular liver disease: multiple biliary hamartoma- report of a case

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Introduction: Multiple biliary hamartomas (MBHs), also known as von Meyenburg's complexes (VMCs), were first described by von Meyenburg in 1918. They are rare benign malformations that consist of multiple well-circumscribed collections of duct-like structures lined by biliary epithelium and surrounded by fibrous stroma. MBH is asymptomatic and usually found incidentally. Its clinical significance is that it may be easily confused with liver metastasis, microabscess and other cystic liver disease.

Case report: A 73-year-old man was admitted to the gastroenterology department of the Cheng-Ching General Hospital with fever and abdominal pain for more than 2 days. The patient had past medical history of hypertension that was controlled by amlodipine (5mg per day for more than 20 years). This time, he suffered from right upper abdominal dull pain since two days before admission. The painful sensation occurred nearly all day, and neither exacerbated nor relieved factors could be traced. There was no nausea, vomiting, or diarrhea but fever up to 39.2 C with chills occurred. Upon admission, physical examination revealed tenderness over right upper quarter of abdomen and positive finding of Murphy's sign. No jaundice was noted. Laboratory data showed leukocytosis with left shift (white blood cell count- 18500/mm³ with segment 94.3%, lymphocyte 1.23%) and elevated C- reactive protein level (CRP-10.2mg/dl). Abdominal sonography was performed and disclosed multiple small hyperechoic nodules, 1-5 mm in diameter, distributed uniformly throughout the liver (figure 1), multiple gall bladder stones and thickness of GB wall. Further investigation with computerized tomography revealed multiple small low-density lesions over both lobes without contrast enhancement (figure 2) and suspicion of cholecystitis. Then antibiotics including cephalosporin and aminoglycoside were administered. MRI and magnetic retrograde cholangiopancreatography showed numerous tiny lesions ranging from 1 to 10 mm diffusely distributed in both lobes. These lesions had low signal intensity on T1-weighted images and increased signal intensity on T2-weighted images. The signal intensity on T2-weighted images was slightly less than that of simple fluid (Figure3). No communication of these lesions and biliary tree was found by MRCP. A surgeon was consulted for laparoscopic cholecystectomy and liver tumor biopsy. During the operation, multiple whitish lesions, 2-3 mm scattered diffusely over liver surface was noted and biopsy and cholecystectomy were done. The pathological report showed multiple biliary channels lined by regular cuboidal epithelium with dense fibrous stroma (Figure 4). A diagnosis of biliary hamartoma was made. The patient was discharged 5 days post-surgically.

Conclusion: biliary hamartomas are unusual benign biliary malformations. Due to asymptomatic, they

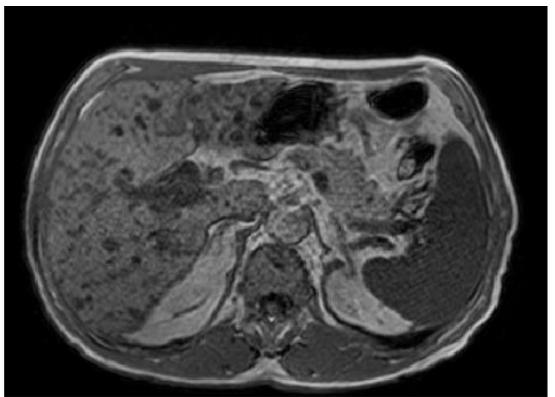
are usually found incidentally and easily misdiagnosed with liver metastases, microabscesses and other cystic lesions. With improvement of the image modality, the diagnostic rate of biliary hamartomas increased. Although their benign innate character, the possibility of malignant transformation to cholangiocarcinoma was still existed.



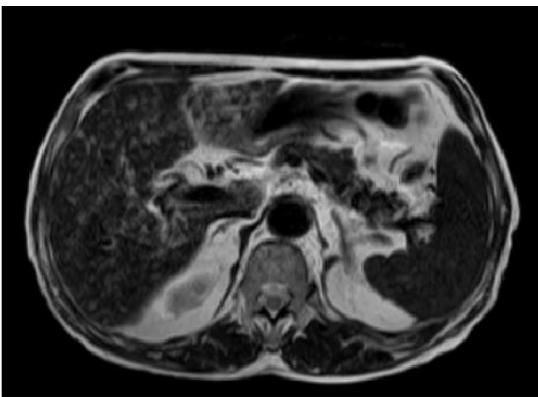
2A



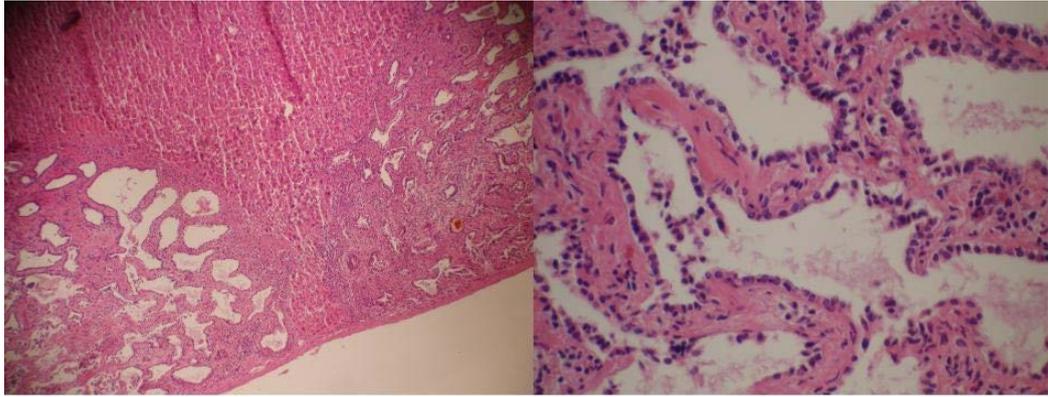
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3A



3B



4A

4B