

A case report of Von Meyenburg Complex (VMCs): Incidental detection in adult dyspnea investigation

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Abstract

Multiple biliary hamartomas, known as 'Von Meyenburg Complexes' (VMCs) are rare mostly benign bile duct malformation which are found incidentally. Most patients have no symptom, non-specific clinical presentation and absent of guidelines management are challenged. Tissue biopsy is performed to differentiate potential malignancy in suspected case. This study presented a case of a patient who had fatigue and dyspnea post covid-19 infection which incidentally found multiple liver cystic lesions, the observation and imaging follow up showed stable lesion without malignant feature.

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Introduction

Multiple biliary hamartomas, known as 'Von Meyenburg Complexes' (VMCs) are rare benign bile duct malformations first described by von Meyenburg in 1918. These are caused by the failure of embryonic remnants of bile duct involution. The prevalence estimated ranges from 0.6%-5.6% from autopsy and around 1% on imaging [1,2]. The VMCs have been reported 0.9% in children [3]. Recently they are increasing report in adults over the age of 35 [1,4]. These variety of cystic lesions of the liver are generally asymptomatic and usually incidental diagnosis on autopsy, intraoperation or imaging work up for other conditions [4,5]. Multiple liver cystic lesions must be differentiated from other lesions such as mimicking liver metastasis [6].

This article is aimed at reporting a case of VMCs present with dyspnea and incidental finding from chest Computed Tomography (CT), diagnosis, follow up and conservative treatment outcome. This case report was approved by the University Research ethics committee (WUEC-25-205-01).

Case presentation

A 49-year-old Thai male patient presented at OPD of School of Medicine, Walailak University Hospital, Nakhon Si Thammarat, Thailand, with fatigue and dyspnea post Covid-19 infection for 2 months. The patient denied previous medical condition or underlying disease. His vital signs were stable. On physical examination found no abnormality was detected. Initial blood work up for Complete Blood Count (CBC), renal function, liver function and thyroid function test were unremarkable (Table 1).

Treatment and follow up

This patient was treated by medication; bronchodilator inhalator for symptomatic relief, molnupiravir (200 mg) 1 tab every 12 hours for 5 days. He was still complaining of fatigue and dyspnea. After failure of medication treatment, the electrocardiography and echocardiography were requested for re-evaluation. The result of that investigation showed normal. The CT chest was performed and reported innumerable Too Small to Characterize (TSTC) lesion scattering both hepatic lobes (Figure 1). The

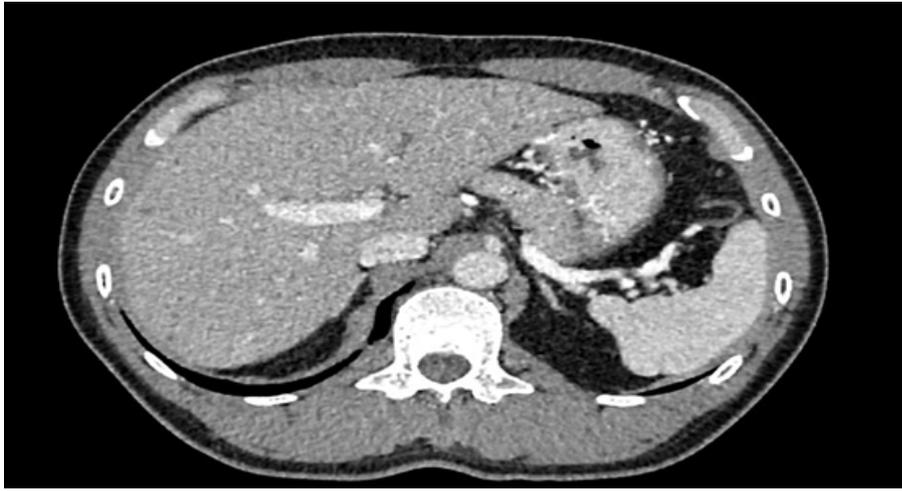


Figure 1: The CT was reported innumerable Too Small to Characterize (TSTC) lesion scattering both hepatic lobes.

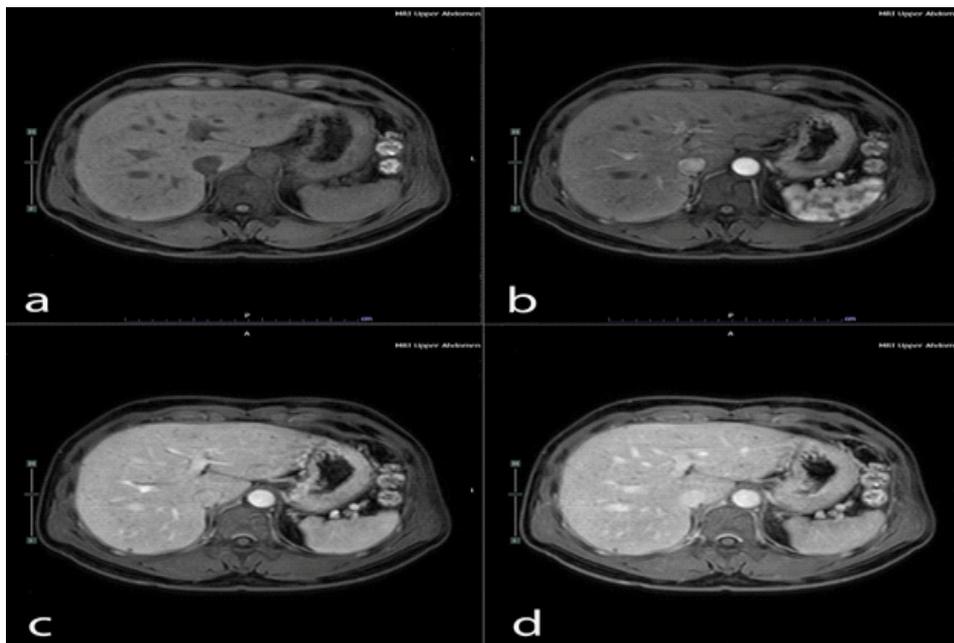


Figure 2: Non-enhancement liver cystic lesion; (A) Pre-gadolinium, (B) Arterial phase, (C) Venous phase, (D) Delay post-gadolinium.

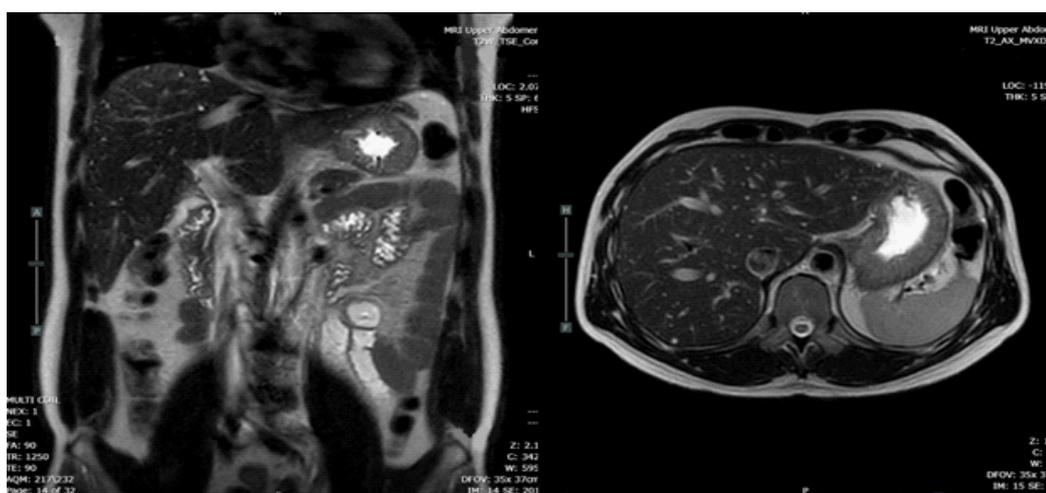


Figure 3: The MRI revealed innumerable cystic lesion in both lobes of liver like a 'starry sky'.

further Magnetic Resonance Imaging (MRI) revealed innumerable cystic lesion in both lobes of liver, (Figure 2). T1W showed a signal lower than the liver parenchyma and T2W showed cystic hyperintensity like a 'starry sky' (Figure 3).

This patient was informed imaging result and advice observed after discussion prognosis based on current clinical evidence. Further clinical and imaging follow up 12-month to date, lesion showed stable without malignant feature on ultrasonography.

Table 1: Complete Blood Count (CBC), renal function, liver function and thyroid function test.

CBC	Result value	Reference	
RBC count	4.48	106 cell/mm ³	3.75-6.54
Hemoglobin	13.7	g/dL	11.5-17.0
Hematocrit	40.4	%	37.0-54.0
MCV	90.2	fl	80.0-100.0
MCH	30.6	pg	27.0-32.0
MCHC	33.9	g/dL	32.0-36.0
RDW	13.2	%	11.0-16.0
WBC count	6490	cell/mm ³	4000-10000
Neutrophil	56	%	40-75
Lymphocyte	36	%	20-45
Monocyte	6	%	10-15
Eosinophil	1	%	0-6
Basophil	1	%	0-1
Platelet count	253000	cell/mm ³	140000-400000
Thyroid function test	Result value	Reference	
Free-T3	3.58	pg/mL	2.02-4.43
Free-T4	1.31	pg/mL	0.93-1.71
TSH	2.76	uIU/mL	0.27-4.20
Renal function test	Result value	Reference	
Blood Urea Nitrogen (BUN)	12	mg/dL	8.4-20.6
Creatinine	0.92	mg/dL	0.73-1.18
eGFR	98	mL/min/1.73m ²	
Liver function test	Result value	Reference	
Aspartate aminotransferase	25	U/L	Nov-34
Alanine aminotransferase	21	U/L	<45

Clinical discussion

Multiple biliary hamartomas or 'Von Meyenburg Complexes' (VMCs) are a variety of benign cystic lesion of liver that consist of focal collections of duct-like structures embedded in a fibrous stroma resulting from ductal plate malformation involving the small interlobular bile ducts [7].

VMCs usually do not cause symptoms and incidental finding with recent noninvasive imaging modalities. Most of biliary hamartoma are composed of small, 5-30 mm disorganized clusters of dilated cystic bile ducts that appear throughout the liver and generally do not communicate with biliary tree [8]. The CT scan of VMCs consists of multiple small, low attenuated areas that do not enhance on contrast injection. MRI of liver and MRCP show low signal intensity on T1-weighted and high signal intensity on T2-weighted images sequences give the liver a 'starry sky' appearance [9-11].

Confirmation of diagnosis VMCs may require liver biopsy. In this patient, asymptomatic bilateral multiple cystic lesions and characteristic imaging finding can help avoid unnecessary invasive interventions. Most of VMCs are benign and usually conservative treatment [12].

Periodic clinical monitoring and determination of CA 19-9 can apply in asymptomatic case, but the use of CA 19-9 and other blood analysis to diagnose malignant transformation should be discouraged because it has low specificity and persistent elevation can present without malignancy [13,14]. In case of alarm symptoms or elevation of tumor marker, perform MRCP with gadolinium [15]. For suspected malignant lesion, consider tissue biopsy to confirm diagnosis. In this case, bilateral multiple cystic lesions of liver, follow up was considered after physician, patient and family discussion about prognosis. Although incidental finding and do not have symptoms, this case should be follow-up, evidence of malignant potential of VMCs has been reported [16,17].

Conclusion

VMCs are rare benign multiple cystic lesions of liver in adult. Treatment usually conservative and follow up. The possibility of malignancy must be discussed and considered as differential diagnosis. Periodic clinical monitor or tissue biopsy to confirm diagnosis should be applied in selected patient.

Declarations

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