SYSTEMIC LUPUS ERYTHEMATOSUS AND MULTIPLE HEPATIC HEMANGIOMAS: A CASE REPORT

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Abstract
Systemic lupus erythematosus (SLE) is a systemic autoimmune disease involving multiple organs, including the liver. In SLE, the best-known involvement of the liver is autoimmune hepatitis. Hepatic hemangioma is the most common benign liver mass in the general population. In most patients, liver hemangioma is incidentally detected during abdominal imaging performed for another reason. Not many studies are available on hepatic hemangioma during SLE. A few case reports have been published in the literature, and the frequency of hepatic hemangioma in SLE was examined in a prospective study. Here, we report a 36-year-old woman diagnosed with SLE based on autoimmune hemolytic anemia, generalized lymphadenopathy, pleural effusion, renal involvement, and serological findings. A mass in the liver was detected on abdominal ultrasonography (USG), and multiple hepatic hemangiomas were detected on magnetic resonance imaging (MRI) of the liver. In this case report, a patient with SLE diagnosed with multiple hepatic hemangiomas was reported, and attention was drawn to the fact that the incidence of hepatic hemangioma in SLE may be increased.

Keywords: Systemic lupus erythematosus; hepatic hemangioma; liver mass

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Key Messages for Research and Practice

• The liver is one of the organs that can be involved (autoimmune hepatitis etc) in SLE patients.
• The incidence of liver hemangioma has increased in the general population. The incidence of liver hemangioma in SLE patients is even more increased compared to the general incidence.
• In patients with SLE, comprehensive prospective studies are needed to reveal the frequency of liver hemangioma and the effect of aging on hemangioma size and course.
Introduction

Hepatic hemangioma is the most frequently seen benign tumor of the liver and is usually detected incidentally during radiologic examinations. Its pathophysiology has not been well elucidated. In studies, the prevalence varies between 0.4% and 20%. The female-to-male ratio is approximately 5:1. In most cases, hemangioma appears as a single lesion, but multiple lesions may also be seen [1-3]. In systemic lupus erythematosus (SLE) patients, autoimmune hepatitis is the best-known form of liver involvement. In addition, fatty liver, cholestasis, chronic persistent hepatitis, nodular regenerative hyperplasia of the liver, and hemangioma may be diagnosed at certain rates [4]. In a prospective ultrasonography (USG) study, the presence of SLE was associated with a 5-fold increased possibility of liver hemangioma [5].

In this case report, we aimed to draw attention to the fact that the incidence of hepatic hemangioma may be increased in SLE by presenting a case with hepatic hemangioma.

Clinical case

A 39-year-old female patient was admitted to the hospital with complaints of leg swelling for a month and shortness of breath for two weeks. The patient had no known history of chronic disease or drug use in her medical history. She was referred after pleuracan insertion due to a massive bilateral pleural effusion detected in another center. In positron emission tomography, increased F-18 FDG involvement was detected in multiple lymph nodes in the left supraclavicular, both axillary, and both inguinal regions, and a lymph node biopsy was found to be reactive. She was admitted to the internal medicine service of our center with a preliminary malignancy diagnosis. Physical examination: fever: 37.8 °C, extensive decrease in lung sounds, pleuranac catheter on the left lung, 2-cm lymph nodes in bilateral axillary and inguinal regions, and bilateral pitting edema. The results of the tests performed during hospitalization in the internal medicine service were as follows: Hb: 8.4 g/dL, leukocyte count 6.14 10^3/U, absolute neutrophil count 4.81 10^3/U, platelet count 245 10^3/U, erythrocyte sedimentation rate 103 mm/hour, C-reactive protein 45 mg/L (0-0.5), ferritin 280 ng/mL, and direct Coombs positive. Biochemical tests resulted as follows: sodium 140 mEq/L, potassium 6.54 K mEq/L, urea nitrogen 46.7 mg/dL, creatinine 1.86 mg/dL, total protein 5.27 g/dL, albumin 2.05 g/dL, aspartate aminotransferase 11 U/L, alanine aminotransferase 5 U/L, lactate dehydrogenase 135 U/L, proteinuria of 5168 mg/day in 24-hour urine and 5-10 leukocytes per area in urine sediment, dysmorphic erythrocytes of 30%. Complement 3 and complement 4 levels were low; antinuclear antibodies were positive at 1/1000-1/3200 (AC-1 nuclear homogeneous); anti ds-DNA was >200.0 IU/mL (19-20). The patient for whom the rheumatology department was consulted was diagnosed with SLE based on autoimmune haemolytic anemia, generalized lymphadenopathy, pleural effusion, renal involvement, and serologic findings; a renal biopsy was performed, and class 4 lupus nephritis was detected; a pulse steroid (1 g/day for 3 days) and 1 g of cyclophosphamide were given intravenously. On abdominal USG, hypoechoic lesions in segments 6 and 2 of the liver parenchyma, the largest of which was 35x26 mm in segment 2, were detected. On magnetic resonance imaging (MRI) of the liver, two nodular-shaped lesions with lobulated contour and intermediate T2 signal intensity, one of which was 3 cm in diameter at the right lobe segment V level and the other 2.5 cm in diameter at the left lobe segment IV level, and several hemangiomas with high signal intensity on T2AGs, the largest of which measured 2 cm in diameter at the segment VIII level, were detected in the liver. The patient was confirmed to have multiple liver hemangiomas with the present findings.

Written informed consent was obtained from the patient for the publication.

Discussion

Liver hemangioma is the most commonly seen solid lesion of the liver, with an incidence of up to 20% [2]. It is difficult to determine the exact incidence of liver hemangioma. Many lesions do not cause symptoms, so a diagnosis cannot be made. They are often detected incidentally [6]. As imaging methods have become more effective and accessible, the rate of detection of hepatic hemangiomas has increased. The prevalence varies depending on the method used. They can be detected in 2-4% of cases through USG, in 5% of cases through computed tomography, and in 7% of autopsy cases [7]. They can occur at any age. The vast majority of hemangiomas remain clinically silent. Very few patients are symptomatic due to the effect of mass, complications, or compression to adjacent structures [2,6].

Although there are case reports describing hepatic hemangiomas in patients with SLE, not many studies have been conducted so far on the frequency. There is an autopsy study and a prospective study on liver hemangioma in SLE [4, 5, 8]. In an autopsy series published in 1992,
the livers of patients with SLE were also evaluated. In this Japanese autopsy study, which included 1468 patients, the rate of fatty liver disease was 11.5% and the rate of hepatic hemangioma was 1.5%. Fatty liver disease was detected in 73% and hepatic hemangioma in 5.7% of the 52 SLE patients in this group. According to this study, the frequency of hepatic hemangioma in patients with SLE was approximately four times higher than that in all patients [4]. In a prospective study in which the frequency of hepatic hemangioma in SLE was evaluated, 35 patients with SLE were compared with 35 healthy controls. The frequency of hemangioma was found to be 54% in the SLE group evaluated through USG and 14% in the control group (p<0.0001). In the relevant study, the authors proposed that SLE increases the possibility of hepatic hemangioma 5-fold and that this may be considered one of the hepatic symptoms of SLE [5].

Hemangiomas are usually solitary, smaller than 5 cm in size, and appear as a well-demarcated lesion that partially collapses during sectioning [2]. In their study, Berzigotti et al. detected solitary hemangiomas in 63% of patients. In this study, the rate of multiple hemangiomas was determined to be 17%, and multiple hemangiomas were associated with a longer duration of SLE [5]. Our patient had multiple liver hemangiomas. However, hemangioma was detected during the diagnosis of SLE. Therefore, we cannot suggest that this was associated with the duration of SLE.

The pathogenesis of liver hemangiomas remains unclear. A vascular malformation is the cause of hemangiomas, which grow in size as the tumor's vessels enlarge. They are more common in women, probably due to the hormonal characteristics of women [6]. The size of hemangiomas may increase over time during pregnancy and estrogen therapy [9]. Berzigotti et al. suggested that the higher number of hemangiomas in people with SLE might be due to angiogenic factors like vascular endothelial growth factor, which is linked to estrogen levels in the blood and disease activity [5].

There are many factors that affect the size of hemangiomas. Some studies examining the natural course of hemangiomas have found that the majority of them remain stable in size, but there are also studies that show that size of liver hemangiomas decrease as age increases [10, 11]. A study in which 211 patients were evaluated showed that hemangiomas tend to increase in size in patients under 40 years of age, while those over 60 years of age tend to decrease in size. The authors attributed the change in size of hemangiomas to the balance between growth factors caused by clotting disorders and growth-inhibiting factors caused by liver fibrosis [11]. With aging, both the clinical features and serologic features of SLE are changing [12]. With aging in the SLE, there is a decrease in phagocytic capacity and production of interferon (IFNa). In addition, there are changes such as NK cell dysfunction, telomere shortening, DNA damage, autophagy and a decrease in B cell count [13, 14]. Many studies have found that the severity of SLE decreases with aging [12]. It is not known how liver hemangiomas change with age in SLE.

**Conclusion**

In conclusion, hepatic hemangioma is the most common solid lesion of the liver in the healthy population. In several studies, it has been shown that the probability of liver hemangioma is increased in patients with SLE compared to a healthy population. The possibility of hemangioma should not be ignored in patients with SLE who are diagnosed with a mass in the liver. More comprehensive studies are needed to clarify whether the presence of SLE is a risk factor for the development of hemangioma. It will also be interesting to know how aging in patients with SLE will make changes in the frequency or size of liver hemangioma.

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**AUTHORS CONTRIBUTION**

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**CONFLICTS OF INTERESTS**

Both authors have completed the ICMJE Disclosure Form (http://www.icmje.org/disclosure-of-interest/; available on request from the corresponding author). Both authors declare that there are no potential conflicts of interest.

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REFERENCES


