



Spontaneous Rupture of Hepatic Hemangioma: Two Case Reports and Literature Review

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Abstract

Hepatic Hemangioma (HH) is congenital vascular malformation, considered the most common benign mesenchymal hepatic tumors, composed of masses of blood vessels that are atypical or irregular in arrangement and size. In most cases, hepatic hemangioma is asymptomatic, and patients have an excellent prognosis. According to previous studies, the size and location of the tumor are correlated with the appearance of symptoms and complications. Cases of spontaneous rupture of hepatic hemangioma have been rarely reported in the literature. In this paper, we described two patients who had spontaneous rupture of hepatic hemangioma in our hospital, and hemangiomas are less than 10 cm in diameter.

Keywords: Hepatic hemangioma; Spontaneous rupture; Surgery

Introduction

Hepatic hemangioma is the most common benign tumor of the liver, and it is often asymptomatic, is usually found incidentally during radiological studies, has a prevalence of approximately 20%, and is more frequent in women. In clinical, cavernous hemangioma is the most common type and most patients have an excellent prognosis because of the benign nature hemangioma [1]. So, most scholars propose that surgery should be restricted to specific situation. Although the reported incidence of hepatic hemangioma with ruptured bleeding was found to be low (1% to 4%), the mortality is rather high. Thus, spontaneous rupture is the most severe complication [2]. To date there are no scientific data correlating the size of the hemangioma with the risk of rupture. In here, we presented two patients with spontaneous rupture of hepatic hemangioma in our hospital, and hemangiomas are less than 10 cm in diameter.

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Case Presentation

Case 1

A 33-years-old woman admitted to our hospital with persistent pain in the upper quadrant of the abdomen, which had occurred suddenly, and pain does not relieve after resting or changing position. She provided a negative history for any medical condition (including hepatitis), recent trauma, or oral contraceptive use. Admission Physical Examination (PE): Temperature (T) 36.5°C, Heart Rate (HR) 75 beats/min, Respiratory Rate (RR) 20 times/min, Blood Pressure (BP) 110/75 mmHg (1 mmHg = 0.133 kPa). Tenderness in the right upper quadrant of the abdomen was noted. Cardiopulmonary function examination showed no obvious abnormalities. Laboratory testing results showed a Hemoglobin (Hb) level of 98 g/L (normal range: 120 g/L to 160 g/L). The rest of the laboratory tests, including liver function and coagulation function are normal. The abdominal enhanced-CT results (Figure 1A): A large round shape with slightly lower confounding density shadow can be seen in the right lobe of the liver. In the venous phase and the delayed phase, the enhancement is obvious, showing a tendency of inward filling, the local capsule is blurred, and a small amount of effusion is seen around the liver. Based on the results, we considered a diagnosis of rupture and hemorrhage of hepatic hemangioma. Considering the threat of further rupture of the hemangioma, the patient underwent an emergency exploratory operation. During the operation, a dark red mass of about 10 cm was seen in the right liver, which was tough in quality. The mass is slightly adhered to the diaphragm and lateral abdominal wall, and a small amount of hemorrhage can be seen around the liver. So hepatic hemangioma resection was performed. The pathological findings revealed rupture hemorrhage of hepatic hemangioma (Figure 1B). The patient recovered well (Figure 1C) and was discharged 5 days after surgery.

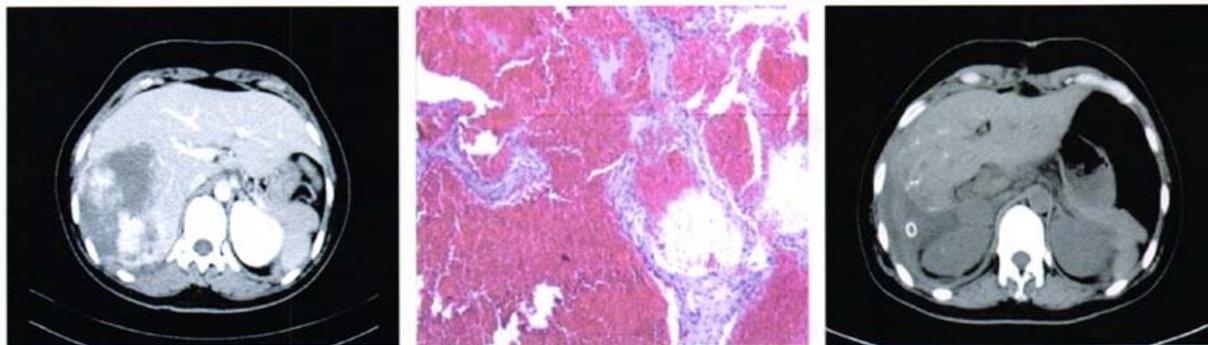


Figure 1: Case 1: A) Preoperative imaging examination, B) Pathological findings, C) Postoperative imaging review.

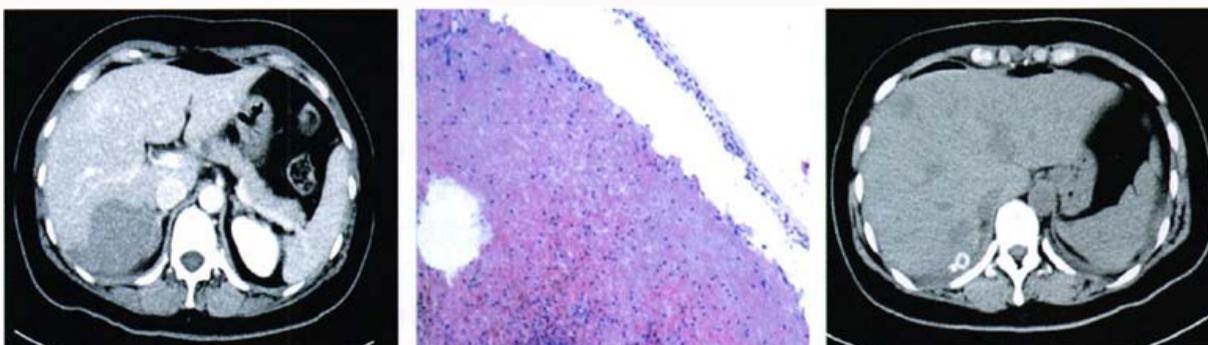


Figure 2: Case 2: A) Preoperative imaging examination, B) Pathological findings, C) Postoperative imaging review.

Case 2

A 36-years-old woman was referred to our hospital with a chief complaint of “sudden right the upper quadrant pain for 14 h”. She provided a negative history for any medical condition (including hepatitis), recent trauma, or oral contraceptive use. PE indicated abdominal pain, especially the upper right abdomen. There was no obvious muscle tension and rebound pain. The blood test results were normal. The enhanced abdominal CT showed a mass in the right liver lobe, which was a patchy progressive enhancement shadow, a fusiform shape was visible in the adjacent liver capsule, and there was a small amount of fluid around the liver (Figure 2A). The patient underwent exploratory laparotomy followed by liver segmentectomy. The pathological findings revealed rupture hemorrhage of hepatic hemangioma (Figure 2B). The postoperative outcome was favorable (Figure 2C).

Discussion

Hepatic hemangioma is the most common benign liver tumor and often found in women, with the majority being cavernous hemangiomas, with little chance of malignant transformation [3]. With the improvement of health conditions and people’s health awareness, more and more patients with hemangioma of the liver have been found. The cause of the disease is not clear. Hepatic hemangiomas grow slowly, and the course of the disease lasts for several years. In general, a hepatic hemangioma with a diameter <4 cm is asymptomatic and prone to be neglected, being discovered only as an incidental imaging finding. Hepatic hemangiomas with diameter >4 cm are regarded as giant hemangiomas and may cause symptoms such as abdominal discomfort and other catastrophic complications including rupture, internal hemorrhage, coagulation disorder, etc.. [4]. Reports on hepatic hemangioma with internal

hemorrhage are rare, causing a lack of understanding and ignoring its serious harmfulness [5]. The incidence of abdominal bleeding caused by ruptured hepatic hemangioma is relatively low (1% to 4%), and the mortality ranges from 60% to 70% [2], and operative mortality rate from this complication of 36.4% [6]. To date there are no scientific data correlating the size of the hemangioma with the risk of rupture. The first case of spontaneous rupture of a hepatic hemangioma was describes by Van Haefen in an autopsy in 1898 [7]. Yamamoto et al. [8] researched 28 cases reported ruptured tumors ranging in size from 3 cm to 25 cm, many of them located on the surface of the liver. In cases where spontaneous rupture occurs, clinical manifestation consists of sudden abdominal pain, and anemia secondary to a hemoperitoneum. Disseminated Intravascular Coagulopathy (DIC) may also occur. Spontaneous rupture of hepatic hemangioma is challenging because it is considered a life-threatening situation [9]. Bleeding of spontaneous rupture is a severe complication in liver disease, as its clinical signs are not usually specific. Conservative treatment runs the risk of hypovolemic shock, and emergent hepatic resection should be preferred, but high operative mortality [10,11]. Therefore, for patients with clinical symptoms, intense surgery demands, and high-risk complications should be actively treated.

Conclusion

At present, a poor knowledge of the natural history justifies the observation management for all asymptomatic hemangiomas. The patients in here might have died unexpectedly when their hemangioma ruptured into abdominal cavity without surgical treatment. In conclusion, surgery is mandatory for ruptured or bleeding hepatic hemangiomas. We hope that our cases will attract more attention to this complication in clinical work. It requires us to reconsider indications for surgery.

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