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CASE REPORT

Salvage of ineffective transarterial embolization with liver resection in a patient with repeated hepatic angiosarcoma bleeding

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Abstract Rupture with internal bleeding is not an uncommon complication of hepatic angiosarcomas that causes high morbidity and mortality. Transarterial embolization (TAE) may be effective for the temporary hemostasis of ruptured tumors. However, the blood supply of an angiosarcoma can also originate in the portal vein, and repeated internal bleeding may stem from this source. Elective liver resection following initial TAE is the standard curative treatment and is the only effective method for preventing further internal bleeding. High-risk liver resection is the only other choice for rescuing patients with fatal bleeding that cannot be controlled by TAE. We report a 66-year-old female patient with angiosarcoma who presented with repeated bleeding and developed hemorrhagic shock after the first episode of TAE. Following two more episodes of TAE, the patient developed internal bleeding with shock, but angiography revealed no arterial bleeding. Bleeding from the portal vein was highly suspected. To prevent further recurrent bleeding, salvage liver resection was subsequently performed under conditions of poor liver reserve. During the 4-month follow-up period until the patient died, no recurrent bleeding was noted. In conclusion, TAE is only a temporary treatment for internal bleeding from a ruptured hepatic angiosarcoma. Recurrent, potentially fatal bleeding may be caused by supplemental vascularization of the portal vein. Surgical tumor resection is necessary for the curative treatment of hepatic angiosarcomas and the prevention of repeated internal bleeding.

Conflict of interest: The authors declare no conflict of interest.

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1. Introduction

Angiosarcoma is a rare type of liver cancer that represents only 2% of primary hepatic malignancies and ~4% of all angiosarcomas of different origins.[1] The spontaneous rupture of a hepatic angiosarcoma may induce severe intra-abdominal bleeding and is associated with higher morbidity and mortality.[2,3] Transarterial embolization (TAE) may be effective in obtaining hemostasis in most hepatic malignancies. In most reported cases, TAE has been performed as the initial treatment. However, ruptured tumors may induce recurrent internal bleeding and cannot be accessed by TAE because they are supplemented by multiple blood sources.[2,4] Following TAE with hepatectomy is the only definite curative treatment available for hepatic angiosarcomas; moreover, it can prevent repeated, potentially fatal internal bleeding. We present a case of ruptured hepatic angiosarcoma treated by TAE without a subsequent surgical resection. We identified subsequent recurrent bleeding that was caused by an angiosarcoma and could not be controlled by TAE. Successful but risky liver resection was performed as a salvage treatment to achieve hemostasis.

2. Case Report

The patient was a 66-year-old woman without previous liver cirrhosis or any viral hepatitis infection. She worked in a badminton racket factory where she operated lathes and recycling planes. The patient had several medical conditions, including rheumatic arthritis, diabetes mellitus, and hypertension, which were controlled by regular medication. She developed dull pain in the right upper quadrant and was referred to a nearby hospital. Medical personnel noted that the patient was in shock, and she was treated with fluid resuscitation before being transferred to our hospital.

The patient had experienced three similar episodes within the previous 2 months. Initially, she was referred to a nearby hospital with similar presentation and a ruptured liver tumor was highly suspected. TAE was performed to stop the bleeding. Upon admission, according to the patient, a following tumor resection was not performed because of the high operative risk. In the following month, she experienced two similar episodes. Abdominal computed tomography (CT) revealed one heterogeneous tumor in the posterior segment, hemoperitoneum, and contrast extravasation (Figures 1A and 2A). Two angiographies for TAE showed no active arterial bleeding (Figures 1B and 2B). Conservative treatment was applied in both episodes.

On arrival, her vital signs were stable. Laboratory data showed normal hemoglobin levels, low platelet counts (66,000/μL), elevated international normalized ratio (1.29), and elevated total bilirubin (1.9 mg/dL). A triphase abdominal CT was applied. In the arterial phase (Figure 3A), a single 16-cm tumor over the right lobe with hemoperitoneum was noted but no contrast extravasation was identified. In the portal vein phase (Figure 3B), some contrast extravasations were observed in the tumor. Bleeding from the portal vein was highly suspected.

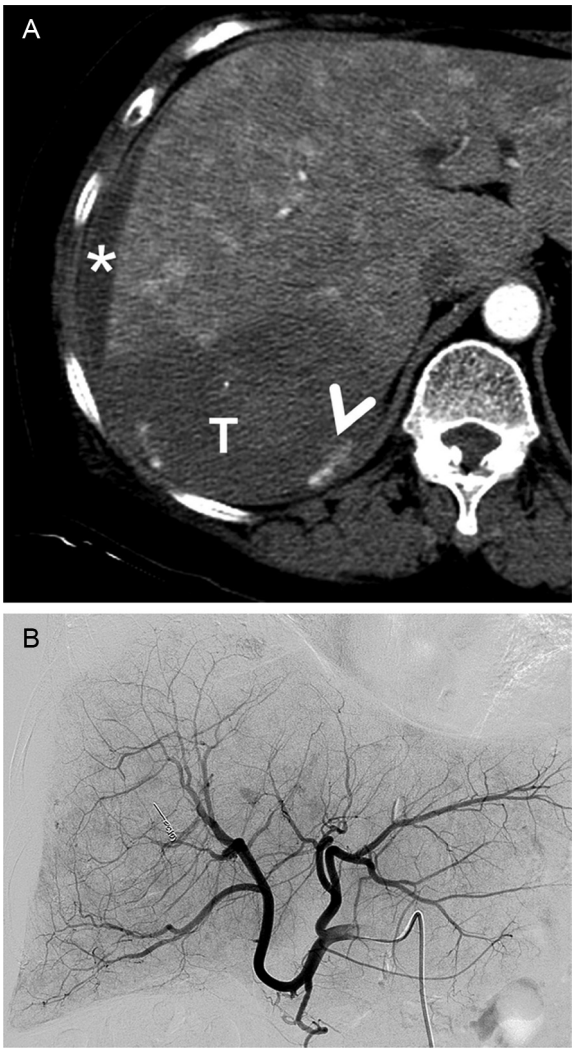

Figure 1 (A) Second bleeding episode: two-phase contrast computed tomography showing a hypodense hepatic tumor with hemoperitoneum and extravasation. (B) Second bleeding episode: angiogram showing previous transarterial embolization coil and no extravasation. Arrow head = extravasation; Asterisk = hemoperitoneum; T = tumor.

Additionally, multiple smaller nodules were observed in the right lobe of the liver. After the examination, her vital signs stabilized with resuscitation and the use of hemostatic medication; hence, no angiography was performed. The preoperative indocyanine green (ICGR-15) test administered after admission was > 30%. The patient was informed of the high risk involved in liver resection, and hepatectomy including Segments 6–8 was performed 1 week after initial presentation to prevent recurrent tumor bleeding. During the operation, we observed a single huge, pulsating tumor occupying the right lobe of the liver and presenting with high vascularity. After the Pringle maneuver was applied to the main portal vein, the tumor underwent shrinkage and bleeding decreased. The resected specimen consisted of a 16 cm × 8.5 cm × 7.2 cm mass. The morphological image revealed a ruptured hepatic angiosarcoma. The resection margin was tumor free.