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CASE REPORT

Improved Survival After Resection for Ruptured Hepatic Angiosarcoma

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Abstract
Hepatic angiosarcoma is rare and accounts for 2% of all primary liver malignancies. Most patients die within 6 months from the time of diagnosis. Surgical resection is the only radical treatment, with a median survival time of around 1.5 years. However, the role of surgery for ruptured tumors has not been determined. A 49-year-old gentleman treated of ruptured hepatic angiosarcoma treated with surgical resection who survived for 11 months is reported here.

Keywords Ruptured tumor · Hepatic angiosarcoma · Surgery · Case report

Introduction

Hepatic angiosarcoma (HS) occurs in up to 2% of all primary liver malignancies and has been considered a very aggressive tumor [1]. Thorotrast, arsenic, androgen, and radium are thought to be etiologic factors, but 75% of cases are undetermined. Symptoms are nonspecific; it is often confused with other liver tumors; thus, the diagnosis of HS is difficult. The overall survival is very poor, most patients die after 6 months of diagnosis [1]. Patient with rupture tumor is even worse, with some figures of < 2 months after embolization [2]. Surgical resection remains the mainstay of treatment, which can significantly prolong survival. However, there is not any published case of prolonged survival after surgical resection for ruptured hepatic angiosarcoma.

Case Report

A 49-year-old man came to the hospital with the main complaint of pain in the right hypochondriac region for 1 week. He had a history of hepatitis B and hemangioma for 1 year, with no specific treatment. Clinical examination was nothing special. He had no anemia, a platelet count of 97 T/L, not suggesting Kasabach-Merritt syndrome. The laboratory results showed a normal liver function and tumor marker. In the computed tomography (CT) scan, we detected free fluid in the right upper quadrant. Two heterogeneous, hypodense mass (diameter of 4 and 7 cm) in the right parenchyma (Fig. 1) was spotted, not excluding malignant tumor. No biopsy was taken due to the risk of uncontrolled bleeding.

A laparotomy using a J-shape incision was performed on the next day with the aim to resect the tumor. There was a serosanguinous fluid collection in the sub-hepatic pouch. The gallbladder was normal. The liver had micro-nodular cirrhosis. The 8-cm hemangioma-like tumor was visible in segments VI and VII. Segments V, VI, and VII also had diffuse lesions (Fig. 2). We choose tumorectomy as the tumor is thought to be a hemangioma. The liver was resected using harmonic scalpel; the gallbladder was also resected.

The intra-operative blood loss was 100 ml. Macroscopic examination showed infiltrative masses. In the microscopy, the lesion type was sinusoidal and vascular spaces with irregular nuclei. The final pathological results revealed

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highly aggressive angiosarcoma (stage pT4 – TNM 8th edition), resection margin > 1 cm. Immunohistochemistry showed positivity in CD31 and ERG staining (Fig. 3). The patient was discharged after 16 days. Follow-up was taken every 3 months, showing no sign of recurrence. He underwent 4 cycles of chemotherapy 7 months later but then died 3 months later due to organ failure.

Discussion

Four main patterns for HS imaging include multinodular, a single dominant mass, mixed patterns of a dominating mass with smaller nodules, or infiltrating micro-nodular tumor. However, these characteristics could not determine HS since it has the same characteristics as other hypervascular tumors, such as hemangioma. And, liver biopsy is considered the only gold standard; however, there is controversy about liver biopsy and the risk of bleeding [3].

No case series analysis has been published. Radiotherapy provides no survival benefit for HS since the tumor is radio-resistant; meanwhile, chemotherapy is still a matter of debate [1, 3]. In addition, liver transplantation is an unfeasible treatment due to its high recurrent rate [4]. Most patients die within 6 months due to advanced stage, liver failure, and hemorrhage. Surgical resection is still the only radical treatment, with a median survival time of 17–19 months [5]. The prognosis for the patient with rupture tumors is even

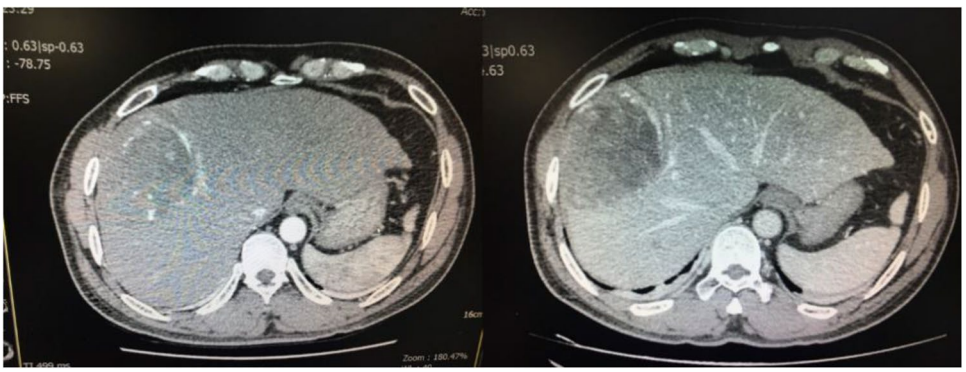

Fig. 1 CT scan showed two large right-sided tumors

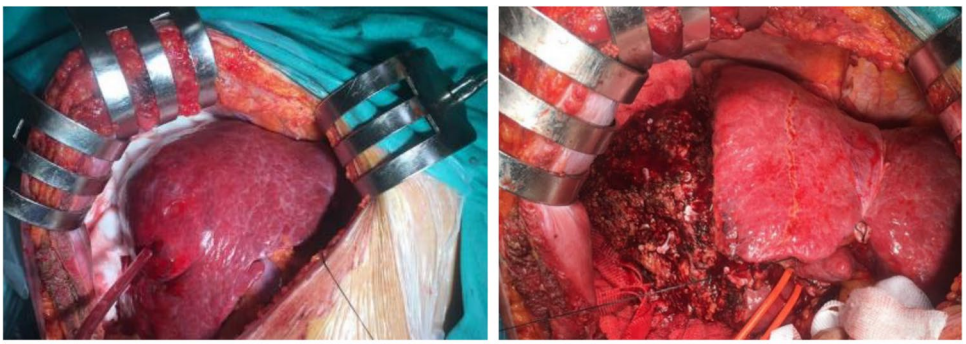

Fig. 2 Intra-operative lesion of angiosarcoma (**a** before resection, **b** after resection)

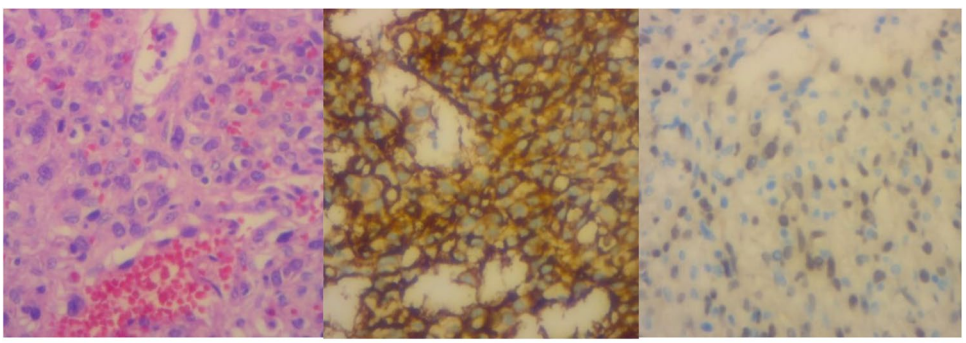

Fig. 3 Immunohistochemistry of angiosarcoma (**a** HE stain, **b** CD31, **c** ERG)

worse (< 2 months), due to tumor cell spread and peritoneal tumor seeding [2]. To some extent, this case demonstrated that survival may be prolonged after surgical resection for a ruptured tumor. Some articles described ruptured tumors treated by embolization, but this method could not control the tumor progression [6, 7]. Meanwhile, the same report showed better survival after surgery, but survival time had not been mentioned [8].

Conclusion

Despite the risk of aggressiveness and peritoneal metastasis, we believe that patients with ruptured tumors might benefit from surgery. Further studies with a larger study population should be conducted to prove the efficacy of surgery in ruptured tumors. The datasets in the current study are available from the corresponding author on request.

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Author Contribution Lan Thi Nguyen: perform the surgery, write the manuscript.

Dang Hai Do: assist the operation, collect the data, write the manuscript.

An Duc Thai: collect the data, write the manuscript.

Hoa Thi Nguyen: do the anesthesia, collect the data.

Declarations

Informed Consent Informed consent was obtained from the patient and his relatives included in the study.

Conflict of Interest The authors declare no competing interests.

References

1. Chaudhary P, Bhadana U, Singh RA, Ahuja A (2015) Primary hepatic angiosarcoma. Eur J Surg Oncol 41:1137–1143
2. Matthaei H, Boelke E, Eisenberger CF, Alldinger I, Krieg A, Schmelzle M, Poremba C, Schellhammer F, Knoefel WT, Budach W, Peiper M (2007) Interdisciplinary treatment of primary hepatic angiosarcoma: emergency tumor embolization followed by elective surgery. Eur J Med Res 12:591–594
3. Zheng YW, Zhang XW, Zhang JL, Hui ZZ, Du WJ, Li RM, Ren XB (2014) Primary hepatic angiosarcoma and potential treatment options. J Gastroenterol Hepatol 29:906–911
4. Maluf D, Cotterell A, Clark B, Stravitz T, Kauffman HM, Fisher RA (2005) Hepatic angiosarcoma and liver transplantation: case report and literature review. Transplant Proc 37:2195–2199
5. Tripke V, Heinrich S, Huber T, Mittler J, Hoppe-Lotichius M, Straub BK, Lang H (2019) Surgical therapy of primary hepatic angiosarcoma. BMC Surg 19:5
6. Leowardi C, Hormann Y, Hinz U, Wente MN, Hallscheidt P, Flechtenmacher C, Buchler MW, Friess H, Schwarzbach MHM (2006) Ruptured angiosarcoma of the liver treated by emergency catheter-directed embolization. World J Gastroenterol 12:804–808
7. Kim B, Reardon R, Cross J, Fehlberg T, Allanson B, Punch GJ (2021) Case report: Haemoperitoneum secondary to acute rupture of primary hepatic angiosarcoma. Int J Surg Case Rep 84:106090
8. Chien C-Y, Hwang C-c, Yeh C-n, Chen H-y, Wu J-T, Chan S-C, Lin C-L, Yen C-l, Wang W-y, Chiang K-C (2012) Liver angiosarcoma, a rare liver malignancy, presented with intraabdominal bleeding due to rupture- a case report. World J Surg Oncol 10:23