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Case Report

Bland embolization of a ruptured hepatoblastoma with massive intraperitoneal hemorrhage ☆

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ABSTRACT

Purpose: Hepatoblastoma is the most common primary neoplasm of the liver in the pediatric population, usually diagnosed during the first 5 years of life. Patients with large or peripheral hepatoblastomas are at risk for rupture and peritoneal hemorrhage. Image-guided, minimally invasive interventions are offered for controlling hemorrhage. Case presentation: We present a 2-year-old female with an 11.8 cm hepatoblastoma in the right hepatic lobe involving segment 4A, who developed hemodynamic instability on day 8 of induction chemotherapy. Imaging revealed intraperitoneal hemorrhage secondary to her ruptured hepatoblastoma. The patient was successfully treated by celiac artery angiogram and transarterial bland embolization. Conclusion: Transarterial bland embolization of large hepatoblastomas may control and even prevent intraperitoneal/intracapsular hemorrhage, and may also enhance the efficacy of systematic chemotherapy in the pediatric patients with advanced hepatoblastoma.

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Introduction

Hepatoblastoma is the most common primary neoplasm of the liver in the pediatric population, typically diagnosed during the first 5 years of life [1]. While hepatoblastoma was associated with less than 30% survival prior to introduction of new chemotherapy regimens, patient survival has significantly improved up to 80% due to advances in imaging, improved surgical techniques and liver transplantation, and improved chemotherapy [2].

Rupture with massive intraperitoneal hemorrhage is an infrequently reported life-threatening complication of hepatoblastoma [3]. Spontaneous rupture and hemorrhage

☆ Patient consent: An informed consent was obtained from the parents for angiogram and possible embolization.

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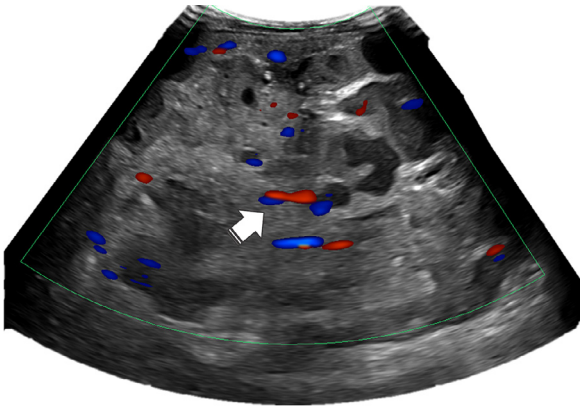

Fig. 1 – Color Doppler ultrasound of liver demonstrates the hepatoblastoma, with scattered internal blood flow (arrow).

are considered poor prognostic factors [4]. Chemotherapy could also interrupt the blood supply to the tumor capsule or surrounding tissue of the tumor, and result in hemorrhage [5].

Surgical intervention for controlling hemorrhage in such an emergency setting is life-saving, however, surgical resection of ruptured hepatoblastoma could result in poor surgical outcome. In modern practice, hemostasis can be achieved by transcatheter arterial embolization (TAE) [6,7].

Herein, we report such a circumstance, a pediatric case of a large hepatoblastoma presenting with massive intraperitoneal hemorrhage following chemotherapy, successfully treated by TAE.

Case presentation

A 2-year-old female with a past medical history of patent ductus arteriosus status post repair presented with an 11.8 cm hepatoblastoma in the right hepatic lobe involving segment 4A. On day 8 of induction chemotherapy, the patient demonstrated clinical signs of decompensation, evidenced by an acute decrease in her hematocrit levels, as well as hemodynamic instability despite transfusion of 2 units of packed red blood cells. On physical examination, her heart rate was approximately 160 bpm and mean arterial pressure was 63 mmHg. Ultrasound of the liver was obtained and again demonstrated the hepatoblastoma, with scattered internal blood flow (Fig. 1). Follow-up computed tomography (CT) angiogram of the abdomen revealed active extravasation into the hepatoblastoma and peritoneum (Fig. 2). Interventional radiology was consulted for urgent intervention and the decision was made to proceed with angiogram and possible embolization.

The right common femoral artery was accessed using a 21-g micropuncture set under ultrasound guidance. This was exchanged for a 3 French vascular sheath. A combination of a high-flow Renegade microcatheter and a 0.016-inch Fathom microwire (Boston Scientific, Marlborough, MA) was used to catheterize the celiac trunk and the proper hepatic artery. Digital subtraction angiogram of both the celiac trunk and proper hepatic arteries demonstrated active extravasation (Fig. 3A). One vial of 100-300 um Embospheres (Merit Medical Systems, Inc., South Jordan, UT), followed by 1 vial of 300-500 um Embospheres was injected, followed by 3 cc of Gelfoam slurry (Pfizer Pharmaceuticals, New York NY) until the bleeding vessels were successfully embolized as evidenced by retention of contrast material within the target vessels and cessation of tumor blush (Fig. 3,B).

Fig. 2 – CT angiogram of the abdomen shows active extravasation into the hepatoblastoma and peritoneum (arrow).

Axial and coronal reformatted contrast-enhanced CT images of the abdomen obtained 4 months postembolization showed complete necrosis of the hepatoblastoma (Fig. 4A and B). At follow-up clinic visit 7 months later, the patient remained clinically stable and endorsed no symptoms. Follow-up CT images at that time demonstrated no imaging evidence of residual hepatoblastoma.

Discussion

This case demonstrated the ability of transarterial bland embolization to successfully treat a ruptured hepatoblastoma. Regardless of intracapsular and intraperitoneal hemorrhage, spontaneous rupture of hepatoblastoma is considered an independent high-risk factor and poor prognostic factor [8]. Chemotherapy was the principal treatment modality in our case, and the tumoral rupture began after initiation of chemotherapy induction. Chemotherapy could induce tumor cell necrosis, which may alter the blood flow to the tumor capsule or surrounding tissue of the tumor, resulting in hemorrhage. This may have been the underlying etiology

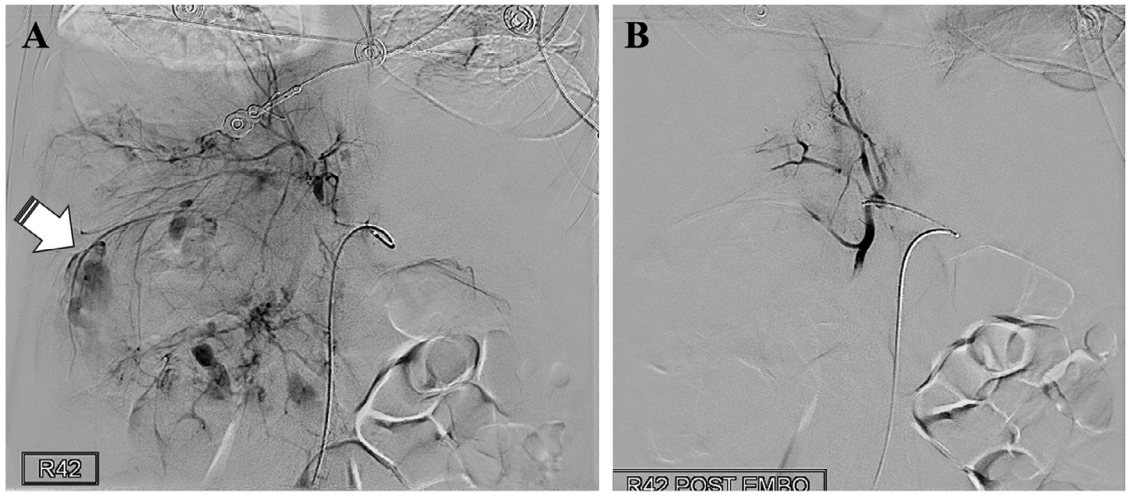

Fig. 3 – (A) Digital subtraction angiogram of the proper hepatic showing multifocal contrast extravasation, consistent with active hemorrhage (arrow). (B) Postembolization digital subtraction angiogram demonstrates resolution of active extravasation.

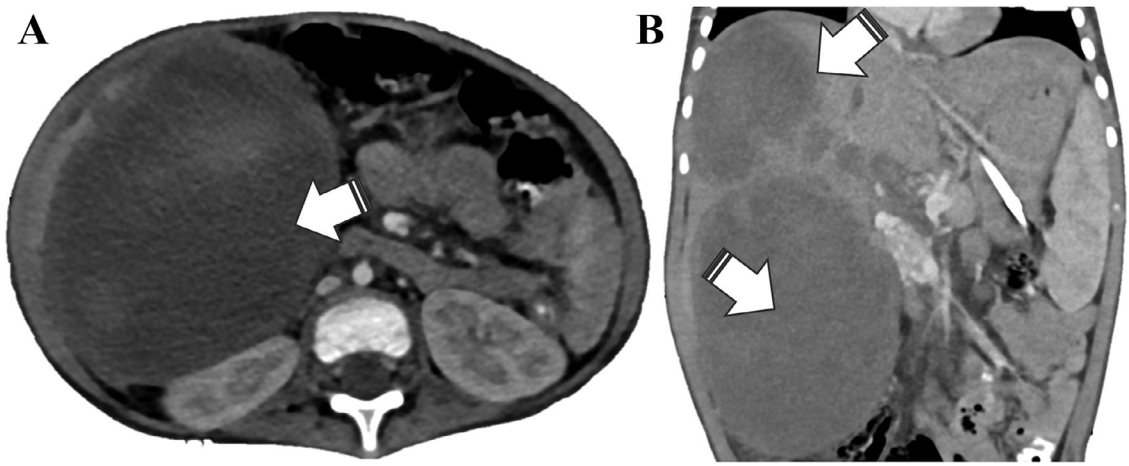

Fig. 4 – Axial (A) and coronal (B) views of follow-up contrast-enhanced CT abdomen demonstrates extensive necrosis (arrow) of the hepatoblastoma.

in our case given the rupture began during chemotherapy induction. Nonetheless, transarterial bland embolization of this advanced hepatoblastoma definitively treated the intraperitoneal/intracapsular hemorrhage, and may have enhanced the efficacy of systematic chemotherapy.

Surgical intervention for controlling hemorrhage in such an emergency setting is life-saving, however, surgical resection of ruptured hepatoblastoma could result in poor surgical outcome. In modern practice, accepted treatment for managing a ruptured hepatoblastoma is TAE, which can then be followed by chemotherapy, surgery, and/or postoperative chemotherapy [6,7]. TAE allows for elective obstruction of arterial flow via the use of various embolic materials. The use of Gelfoam, histoacryl, or microcoils has all been described, and the choice of which depends on various factors, such as the diameter and characteristics of the target artery [6]. Physiologically, the liver parenchyma is supplied from 2 vessels, the hepatic artery and portal vein. However, primary or metastatic neoplasms of liver are mainly supplied by the hepatic artery, with hepatoblastoma being no exception. Therefore, we expected TAE would be effective in occluding the feeding artery of the hepatic tumor, as has been previously described [6], which is what occurred in this case. An additional benefit to TAE is the possibility for expedited tumor shrinkage for patients with advanced hepatoblastoma on chemotherapy who do not respond to systemic chemotherapy [7].

Transarterial chemoembolization (TACE) is another proven approach to selectively treat hepatoblastoma and has been used as an alternative treatment option to systemic chemotherapy [9]. The efficacy of TACE on hepatoblastoma is similar to systemic chemotherapy, but less toxic. TACE is an effective procedure for treating pediatric liver neoplasm. However, TACE is challenging in the pediatric population due to the need for general anesthesia and the small size of both

the patient's overall anatomy and target vessels [10]. The findings of this single case report are encouraging, however, additional clinical trials are required to validate efficacy of TACE for hepatoblastoma.

Hepatoblastomas are common pediatric tumors that can spontaneously rupture, causing massive hemorrhage. In this case of ruptured hepatoblastoma with massive intra-tumoral and intraperitoneal hemorrhage, we demonstrate the possibility for bland TAE to effectively control bleeding and treat the underlying tumor.

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