CASE REPORT

Non-traumatic spontaneous ruptured hepatocellular adenoma after selective transcatheter arterial chemo-embolization for reduction of tumor mass: A case report

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ABSTRACT

It is important to detect hepatic adenomas, which are generally well-confined lesions, as urgent treatment is required due to the risk of bleeding and conversion to malignancy. Dimensions can vary between 1 and 15 cm, and the risk of hemorrhage is higher in large and multiple-focus adenomas. The case is here presented of a patient with spontaneous ruptured liver adenoma after selective transcatheter arterial chemo-embolization for reduction of tumor mass, with discussion of the clinical, computed tomography, angiographic findings, and treatment strategies.

Keywords: Chemotherapeutic embolization; bleeding; hepatic adenoma; liver neoplasms.

INTRODUCTION

Hepatic adenoma (HCA) is typically a solitary benign liver lesion, seen mostly in young women using oral contraceptives. It may be complicated by spontaneous intratumoral bleeding, with or without rupture into the abdominal cavity. Malignant transformation may develop, which has led to the advice to resect lesions >5 cm.[1,2] Selective transcatheter arterial chemo-embolization (TACE) has also been described as an important and safe treatment for large HCAs in an attempt to reduce the tumor mass.[3] In general, an asymptomatic adenoma, it is often detected coincidentally in abdominal examinations performed for different reasons. Tumor rupture is not expected in the early period after the TACE treatment. With the effect of the mass, symptomatic lesions can cause right upper quadrant pain. However, spontaneous rupture, which is one of the important risks, and acute abdominal pain, hypotension, and even loss of life when hemorrhage develops, has been reported.[4] It is important to identify the bleeding point in the tumor with radiological imaging using ultrasonography (USG) and computed tomography (CT) imaging in the emergency department. CT is the most useful imaging modality for diagnosis. Surgery, arterial embolization, or conservative therapy are the treatment options in ruptured HCA patients and are performed according to the status of the disease.

The case is here presented of a 32-year-old female with HCA who had undergone TACE and was then admitted to hospital because of non-traumatic abdominal pain after 48 h. To the best of our knowledge, there has been no previous report in the literature of a patient who developed rupture in the early period after TACE. Consent was obtained from the patient for TACE and the surgical procedure and publication of this article. Our experience is reported here together with the imaging findings.

CASE REPORT

A large hepatic mass was detected incidentally on ultrasound examination of a 32-year-old female patient with no history of oral contraceptive use. The patient had no history of viral hepatitis and laboratory findings were negative for viral markers.

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On the CT scan, arterial-venous and 5th-min venous phase demonstrated a large and irregular hypodense mass in segment 7–8 (Fig. 1). On the magnetic resonance imaging scan, a large mass lesion was detected with heterogeneous enhancement, showing as mild T1AS hypointense and T2AS hyperintense. USG-guided core biopsy of the mass in segment 7–8 was performed twice using an 18-gauge biopsy gun. Histopathological examination showed that the mass was consistent with HCA. The patient treatment plan was formed by a hepatobiliary consultation group, consisting of experienced hepatobiliary surgeons, a gastroenterologist, and an interventional radiologist. The non-contrast CT scan volumetric calculation was consistent with HCA (Fig. 1). Tumor size reduction was planned after TACE and surgical resection if necessary. Endovascular embolization was then performed in the hospital. Selective angiography showed the tumor to

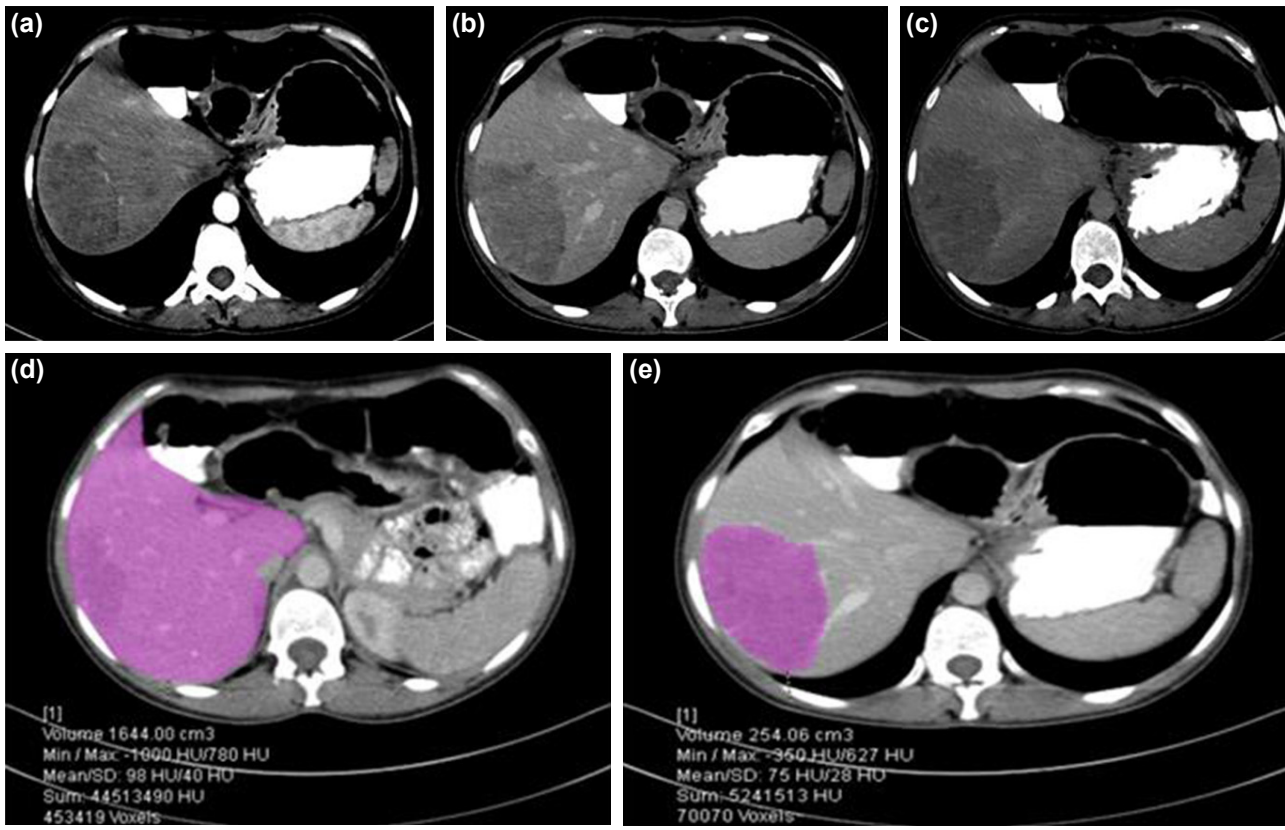

Figure 1. (a-c) Computed tomography (CT) scans in the arterial-venous and 5th-min venous phase show an irregular hypodense large mass in segment 7–8. **(d, e)** Non-contrast CT scans volumetric calculation of HCA.

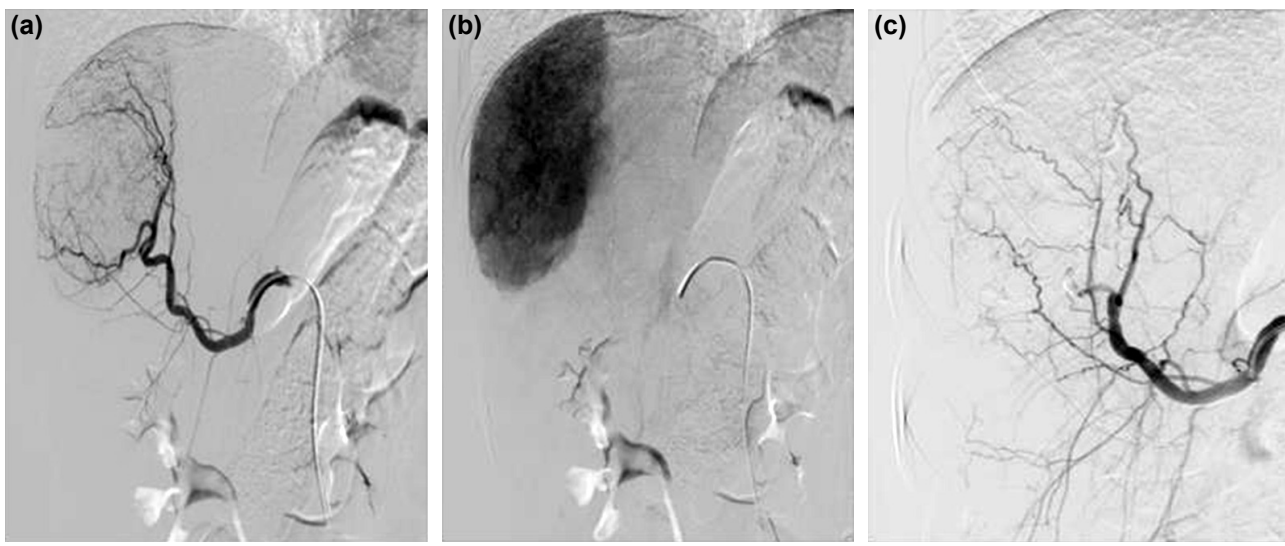

Figure 2. (a) Hepatic arteriography shows numerous hypervascular masses in the right hepatic lobes in the early arterial phase, **(b)** capillary phase of HCA, and **(c)** post-embolization angiography shows occlusion of tumor vessels.

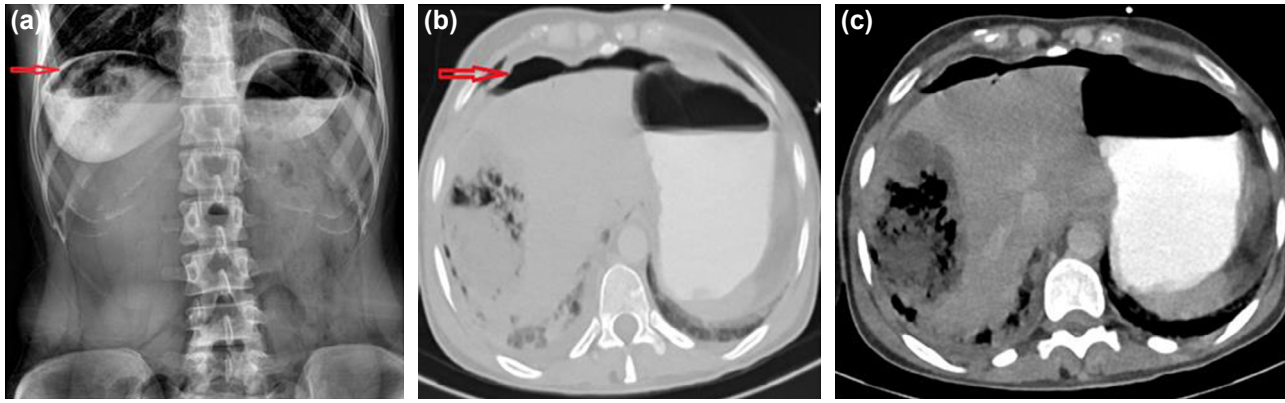

Figure 3. (a, b) Abdomen X-ray and computed tomography (CT) scan show subdiaphragmatic free air (red arrows) and **(c)** contrast-enhanced CT of the liver shows a large non-enhancing lesion in the right lobe, containing a hypodense area of hemorrhage, and necrosis with air.

be highly vascularized. Superselective angiography of the right liver arteries was applied for embolization of the segmental arteries with 5 mg doxorubicin hydrochloride and 100–300 micron particles. The last control angiography showed successful embolization (Fig. 2) for reduction of the HCA and the patient was discharged after 1 day.

Two days later, this patient who had undergone TACE treatment for HCA was re-admitted to hospital because of non-traumatic abdominal pain and abdominal tenderness. Abdominal defense and rebound were determined in the physical examination. Blood pressure was 80/50 mmHg and heart rate was 138 bpm. The laboratory parameters were as follows: Hemoglobin: 7.9 g/dL (12.2–18.1), platelets 127 10^3/uL (60–450), and leukocytes 14.4 10^3/uL (4.3–10.3). Abdominal US revealed a liver mass with air and free fluid. On the abdominal X-ray and CT scan, sub-diaphragmatic free air was observed. Contrast-enhanced CT of the liver showed a large non-enhancing lesion in the right lobe, containing a hypodense area of hemorrhage and necrosis with air (Fig. 3).

Acid sampling was applied under USG guidance. With the determination of hemorrhagic features in macroscopic evaluation, emergency surgery was performed. Bleeding from the mass in the right lobe of the liver was controlled with perihepatic packing tamponade. The bleeding was stopped with the surgical treatment and patient was discharged after 7 weeks.

DISCUSSION

Histologically, HCAs closely resemble normal hepatocytes, consisting of groups of cells, separated by dilated sinusoids. Sinusoids consist of thin-walled capillaries, which do not contain portal blood supply and are fed only from the peripheral arterial system. Therefore, differentiation from hepatocellular carcinoma is difficult because of the arterial hypervascular feature, but due to this feature, TACE can be of benefit. Kupffer cells, of which there are few in adenomas, are lost with functional decrease. In addition, HCA can be distinguished from follicular nodular hyperplasia (FNH) by the absence of the bile duct. HCAs are generally non-lobular lesions with sharply-defined borders and appear as a hyperechogenic lesion on US.[5]

On CT, HCA varies depending on the fatty liver background with density depending on the amount of adipose tissue hepatocytes and Kupffer cells. On pre-contrast CT, portal venous, and late phase scans, the lesion is observed as hypodense compared to the normal liver, with density observed in the arterial phase. On contrast CT, heterogeneous enhancement can be seen depending on the fat and hemorrhage content. However, there is no permanence of contrast due to intratumor arteriovenous shunts.[5,6]

On T1AS MR images, HCA can be hyperintense, isointense, or hypointense, depending on the fat and hepatocyte/Kupffer cell ratio. In addition, hemorrhagic transformation and calcification can be seen to be heterogeneous due to necrosis. On T2AS images, HCA is observed as mildly hyperintense compared to the normal liver. HCAs are followed by hypointensity due to the lack of HCA functional hepatocytes in the 20-min hepatic phase images in hepato-specific contrast application. This feature is an important criterion in differentiating HCA from FNH.[7] Selective TACE may also be used as a treatment to reduce large HCAs.[8] The treatment plan for the present case was HCA size reduction after TACE and surgical resection by hepatobiliary surgeons. Ruptured HCA is known to have quite a high mortality rate and patients with multiple lesions or a lesion >5 cm are at high risk of developing spontaneous bleeding with or without rupture.

The rates of bleeding and rupture are higher with oral contraceptive use compared to patients who do not use oral contraceptives.[8] From a review of the literature, no case could be found of non-traumatic spontaneous rupture of HCA in the early period after TACE. Acute abdominal pain, distension, and shock are symptoms of ruptured HCA. Hemorrhagic shock is a very important risk factor, which increases the mortality rate in these patients.[9]

The present case presented with para-umbilical abdominal tenderness and abdominal distension. Diagnosis can be made from the clinical findings, laboratory parameters, and radiological imaging (USG and CT). In our clinic, US is used with CT examination for diagnosis of acute abdomen, and these methods are readily available in our hospital emergency setting.

Acute therapies include conservative treatment, TACE, and hepatobiliary surgery.[10] TACE is an important option to obtain hemostasis of a ruptured HCA but the patient did not consent to this procedure. The surgical option was selected in this case and adequate hemostasis was obtained with surgery.

HCA rupture is not common 2 days after TACE. The treatment of ruptured HCA is very important as it can be life-threatening. The treatment plan requires a consensus of interventional radiologists and hepatobiliary surgeons. This complex case report can be considered a useful addition to the literature and provides a guideline for the future cases.

Conclusion

Non-traumatic spontaneous ruptured HCA after selective TACE is known to have quite a high mortality rate. The surgical option and endovascular embolization are an effective and safe treatment modality for unstable patients.

Informed Consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

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Conflict of Interest: None declared.

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OLGU SUNUMU - *ÖZ*

Hepatik adenomda tümör boyutu küçültme amaçlı transarteriyel kemoembolizasyon sonrasında gelişen non travmatik spontan rüptür: Olgu sunumu

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Kanama riskleri ve maligniteye dönüşüm potansiyellerinden dolayı Hepatik adenomların önceden tespit etmek önemlidir. Tümör boyutları 1 ile 15 cm arasında değişmekte olup multipl sayıda olanlar ile özellikle büyük boyutlardaki hepatik adenomlarda kanama riski belirgin artmaktadır. Sunduğumuz bu olguda büyük boyutlarda hepatik adenomlu hastada selektif transarteriyel kemoembolizasyon ile tümör boyutunda azalma ile cerrahi rezeksiyon planlanmış ve gelişen nontravmatik spontan tümör rüptür sonrasında klinik, bilgisayarlı tomografi, anjiyografi bulguları yanısıra tedavi stratejileri tartışılmıştır.

Anahtar sözcükler: Hepatik adenom; kanama; karaciğer neoplazm; kemoteropatik embolizasyon.

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