

# Endovascular Treatment of Massive Retroperitoneal Hemorrhage Due to Spontaneous Rupture of Right Adrenal Gland Metastasis that was Secondary to Invasive Hepatocellular Carcinoma: A Case Report<sup>1</sup>

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The reported incidence of spontaneous rupture of primary hepatocellular carcinoma (HCC) is up to 14.5%. However, rupture of the metastatic lesions of HCC is very rare. We describe here a case of massive retroperitoneal hemorrhage due to spontaneous rupture of right adrenal gland metastasis that was secondary to invasive HCC. This was successfully controlled by performing transcatheter arterial embolization (TAE).

**Index words :** Liver neoplasms, metastases

Liver neoplasms, chemotherapeutic embolization

Adrenal gland

Retroperitoneal space

Hepatocellular carcinoma (HCC) is the most common primary malignant tumor of the liver. Extrahepatic metastasis can occur through hematogenous metastasis, lymphatic extension and direct invasion (1).

The incidence of spontaneous rupture of primary HCC has been reported to be up to 14.5% (2, 3). However, rupture of the metastatic lesions of HCC is very rare. We report here on a case of massive retroperitoneal hemorrhage due to spontaneous rupture of right adrenal gland metastasis that was secondary to invasive HCC. This was successfully controlled by transcatheter arterial embolization (TAE).

## Case Report

A 45-year-old man was admitted to our hospital because of severe right upper quadrant pain of an acute onset, abdominal distension and hypovolemic shock.

He was diagnosed with massive HCC in the right lobe of the liver and lung metastasis; the HCC was associated with hepatitis B virus (HBV)-positive hepatitis, which he had been diagnosed with 6 months previously at our hospital. The patient had received transcatheter arterial chemembolization (TACE) three times during the previous 6 months.

The contrast enhanced abdominal CT obtained 4 days before the onset of symptoms shows partial retention of iodized oil in the mass in the posterior segment of the right lobe of the liver with direct invasion to the right adrenal gland (Fig. 1).

On the present admission, the patient's initial blood pressure was 70/40 mmHg and a laboratory test showed a rapid drop of hemoglobin during 4 hours, from 13.2 g/dl to 11.0 g/dl

Unenhanced and enhanced abdomen CT was then performed. The abdominal CT showed a large retroperitoneal hematoma involving the right perirenal and anterior pararenal spaces and it abutted the right adrenal metastasis of the HCC, suggesting rupture. As a result, the duodenum and IVC were displaced to the left side by hematoma (Fig. 2).

Angiography was performed to evaluate for tumor bleeding from an arterial vessel and then to eventually

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embolize it. A 5F Yashiro type catheter (Glidecath, Terumo, Tokyo, Japan) was placed via the right femoral artery approach at the celiac trunk. The aortogram, celiac angiogram and right renal arteriogram showed no tumor staining or extravasations of contrast material. A 5F Yashiro catheter was then placed at the right phrenic artery. The right phrenic angiogram showed tumor staining at the right adrenal gland. A 2F microcatheter (Progreat, Terumo, Tokyo, Japan) was coaxially passed through the 5F Yashiro catheter; the selective right

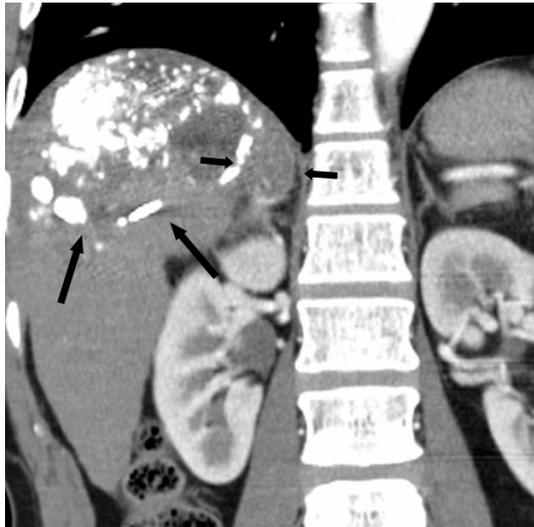


Fig. 1. Coronal reformatted image of the contrast enhanced abdominal CT obtained 4 days before onset of symptoms shows partial retention of iodized oil (arrows) by the mass in the posterior segment of the right lobe of the liver with direct invasion to the right adrenal gland (small arrows).

phrenic artery angiogram then showed tumor staining at the right adrenal gland (Fig. 3A). TACE was initially performed with an infusion of 5 mL of iodized oil (Lipiodol; Laboratoire Andre Guerbet, Aulnay-sous-Bois, France) and 20 mg of doxorubicin hydrochloride (Adriamycin; Pharmacia, Milan, Italy) emulsion into the right phrenic artery. Additional embolization was performed with using gelatin sponge (Cutanplast; Mascia Brunelli, Milan, Italy), which we cut into small pieces and soaked in a solution of 10 mL of nonionic contrast medium (Visipaque; Amersham, Cork, Ireland) and 3 mL of saline solution. The right phrenic artery angiogram after embolization with an emulsion of iodized oil and doxorubicin hydrochloride and gelatin sponge showed retention of the embolized emulsion in tumor cells and there was still residual tumor staining; embolization of the branch of the right phrenic artery was next performed with microcoils (two 2 mm  $\times$  2 cm hilar coils, two 3 mm  $\times$  2 mm tornado coils and six 4 mm  $\times$  2 mm tornado coils, Cook, Bloomington, IN, U.S.A.). The right phrenic artery angiogram obtained after the embolization with additional microcoils showed that the branch of the right phrenic artery was completely occluded, and iodized oil was taken up to the right adrenal gland; tumor staining at the right adrenal gland was no longer seen (Fig. 3B).

After the embolization, the patient's blood pressure was 100/60 mmHg and the vital signs were stable. Abdominal CT was performed again after 6 days and it showed the interval size decreases of the retroperitoneal hematoma



Fig. 2. A, B. The coronal reformatted image (A) and the transverse image (B) of the contrast enhanced abdominal CT show a large retroperitoneal hematoma involving the right perirenal (arrows) and anterior pararenal spaces (arrows); this hematoma abuts the right adrenal metastasis (arrowhead) of the HCC, suggesting rupture. As a result, the duodenum (small arrow) and IVC (arrowhead) were displaced to the left side by the hematoma.

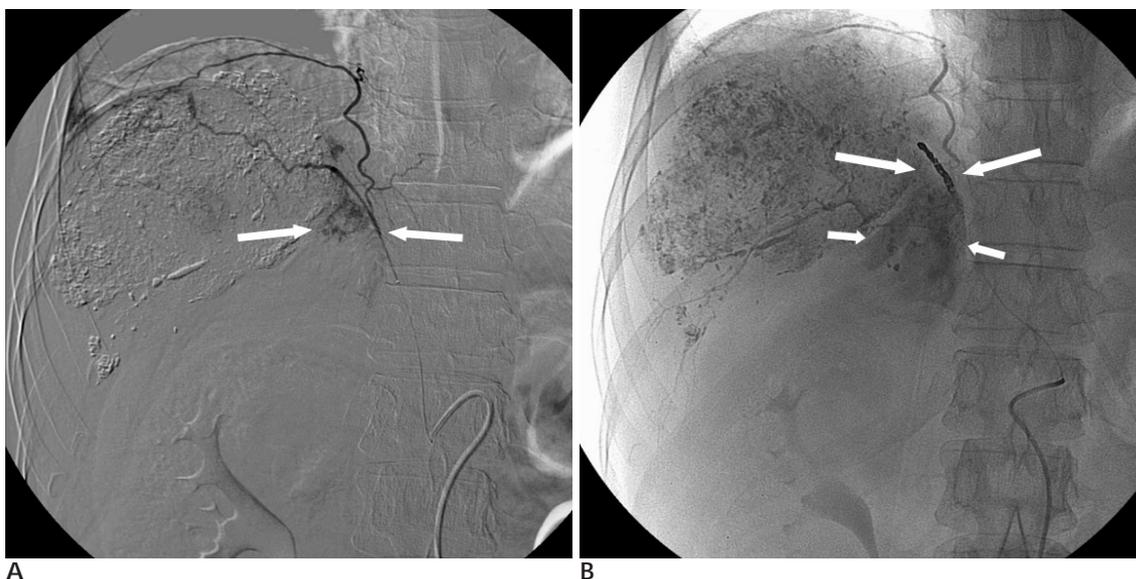


Fig. 3. (A) Selective right phrenic artery angiogram shows tumor staining (arrows) at the right adrenal gland. (B) The right phrenic artery angiogram obtained after transcatheter arterial embolization with an emulsion of iodized oil and doxorubicin hydrochloride, gelatin sponge and microcoils (arrows) shows that the right phrenic artery was completely occluded, iodized oil was taken up by the right adrenal gland (small arrows), and tumor staining at the right adrenal gland was no longer seen.

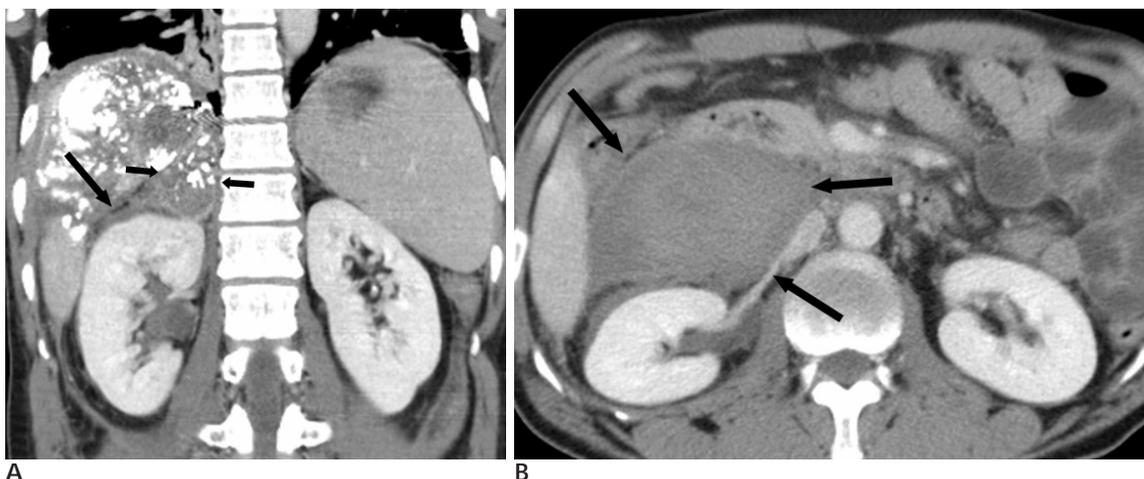


Fig. 4. A, B. The coronal reformatted image (A) and transverse image (B) of the contrast enhanced abdominal CT obtained 6 days after transcatheter arterial embolization shows interval size decreases of the retroperitoneal hematoma (arrows) involving the right perirenal and right anterior pararenal spaces and the uptake of iodized oil in the right adrenal metastasis (arrowheads).

involving the right perirenal and right anterior pararenal spaces, and retention of the iodized oil in the right adrenal metastasis and the embolized microcoils (Fig. 4). The patient was discharged 6 days after the CT scan.

### Discussion

Although the adrenal gland is the second most common site of metastasis from HCC in autopsy cases (4), there has been only one reported case in the Japanese article describing spontaneous rupture of the adrenal metastasis from HCC (5).

The adrenal metastasis in this case was not a hematogenous metastasis, but it was considered as the direct extension to (and engulfing) the right adrenal gland because the normal medial and lateral limbs of the adrenal gland were obliterated and an oval mass was formed (Fig. 1). It is difficult to differentiate rupture of HCC invading the right adrenal fossa from rupture of adrenal metastasis from invasive HCC. For our case, the massive retroperitoneal hemorrhage may have been caused by spontaneous rupture of the right adrenal gland metastasis from the invasive HCC because the retroperitoneal hemorrhage abutted the right adrenal

metastasis of the HCC (Fig. 2A); further, the CT obtained 6 days after TACE showed uptake of iodized oil in the right adrenal metastasis (Fig. 4A)

Each adrenal gland has three sources of arterial blood supply from the inferior phrenic artery, the aorta and the renal arteries (6). Tumor staining was not seen on the aortogram and the right renal arteriogram, and the right phrenic artery was the main feeding artery in this case.

Unruptured adrenal metastasis can be treated by adrenalectomy, TACE or percutaneous ethanol injection (PEIT), according to the clinical features of each individual; these features include the size of the metastatic tumor, whether there is invasion into the IVC, the function of the remaining liver and the existence of intra- and/or nonadrenal extrahepatic lesions (7).

Spontaneous rupture of HCC or metastatic HCC is a critical and life-threatening condition and its prognosis is extremely poor. These patients usually present with a sudden onset of abdominal pain that's accompanied by hypovolemic shock, abdominal distension and massive hemoperitoneum.

The mechanism of ruptured HCC is poorly understood. The postulated mechanisms include rapid growth of tumor and necrosis, rupture by splitting of the overlying normal hepatic parenchyma or erosion of a vessel and occlusion of the hepatic veins by a tumor thrombus (3, 8).

Massive hemorrhage may be one of the causes of hepatic failure in cirrhotic patients with ruptured HCC. Therefore, either emergency surgery or arterial embolization is necessary when faced with this condition. Almost all of these patients are poor surgical candidates because of cirrhosis and extensive tumor replacement of the liver, which carries a high risk of morbidity and mortality. TAE has been reported to be highly effective to achieve hemostasis in patients with ruptured HCC,

and its immediate mortality rate is far less than that of surgery (9, 10). TAE may also be an appropriate treatment even for ruptured adrenal metastasis of HCC.

In summary, we describe here a case of massive retroperitoneal hemorrhage due to spontaneous rupture of right adrenal gland metastasis that was secondary to invasive HCC, and this was successfully controlled by performing TAE.

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