Spontaneous Rupture of Adrenal Metastasis from Hepatocellular Carcinoma

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Rupture of adrenal tumor from various primary origins is a rather rare event. We report here on a ruptured adrenal metastasis from hepatocellular carcinoma, and this ruptured metastasis was observed at the time of the initial diagnosis.

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There have been various reports about cases of spontaneous rupture of adrenal tumor, such as pheochromocytoma, adrenal cortical carcinoma and adrenal myelolipoma. To the best of our knowledge, there has been only one case of adrenal metastatic tumor that caused spontaneous rupture [1]. Because most extrahepatic hepatocellular carcinomas occur in patients with an advanced intrahepatic tumor stage, the first manifestation as spontaneous rupture of adrenal metastasis may be extremely rare [2]. We report here on a patient who presented with spontaneous rupture of a huge adrenal metastasis as the initial manifestation, and the patient had a small primary hepatocellular carcinoma in his noncirrhotic liver.

Case Report

A 56-year-old man presented to the emergency department with suddenly-onset right flank pain that had developed one day previously. The pain was radiating to the epigastric area and the back. The physical examination showed right flank tenderness and right upper quadrant tenderness on palpation. For his past history, he was a chronic alcoholic and had consumed 17g/day alcohol for about thirty years, but the laboratory findings were normal. The tests for hepatitis-B and C antigens were negative. There was no evidence of cirrhosis on the CT images and laboratory findings.

The abdominal CT scan showed a huge mass in the right adrenal gland, measuring about 8.2×7.6×10.0 cm, with adjacent infiltrations. The normal right adrenal gland could not be identified. On the noncontrast CT scan, this adrenal mass consisted of a high-attenuated peripheral portion and a central low attenuated necrotic component that contained foci of central increased density, suggesting a hemorrhagic condition of an adrenal tumor. Dynamic CT scans during the hepatic arterial phase showed the right adrenal mass had poorer peripheral enhancement than that of the liver parenchyma [55HU to 61HU], and there were minimal enhanced foci within the central necrotic portions (Fig. 1A-C). The inferior margin of the adrenal mass was poorly defined, and it appeared to be infiltrative, extending to the anterior and posterior perirenal spaces and retroperitoneum. The inferior capsule of the liver was presumed to be in-
tact. Another small enhancing hepatic nodule was detected on the arterial phase at the subcapsular portion of the fifth liver segment, but the rest of the hepatic parenchyma was relatively unremarkable (Fig. 1D). This hepatic nodule demonstrated peripheral arterial enhancement and isoattenuation, as compared to the liver parenchyma on the equilibrium phase, suggesting hepatocellular carcinoma.

The T2-weighted axial MR images showed a slightly heterogeneous mass in the right adrenal gland (Fig. 2A). There was no detectable fat component within the right adrenal mass on the fat-saturated T1-weighted images (Fig. 2B). The central “whirl” appearing lesion had a hyperintense signal on the T2 weighted images and an isointense signal on the T1-weighted images, which are findings consistent with hyperacute hemorrhage (Fig. 2A, B). The gadolinium-enhanced dynamic MR images also depicted only a thin crescent of arterial enhancement in the anterior peripheral portion, and this portion might have been a viable tumor component in the hepatocellular carcinoma; however, the pathology was not confirmed (Fig. 2C, D).

Palliative excision of the right adrenal gland and hepatic wedge resection at segment V were performed. Microscopic pathologic examination revealed poorly-differentiated carcinoma at segment V and in the right adrenal gland (Fig. 2A). The adjacent infiltrations and retropitoneal hemorrhage suggest rupture of the adrenal tumor (arrowhead).

**Fig. 1.** Abdominal CT scan of a 53-year-old man with right adrenal metastasis from hepatocellular carcinoma.

A. The non-contrast CT scan shows a highly attenuated peripheral portion and hyperdense foci within the central necrotic area in the right adrenal gland (arrow). The adjacent infiltrations and retropitoneal hemorrhage suggest rupture of the adrenal tumor (arrowhead).

B. On the arterial phase, the right adrenal mass is poorly enhanced.

C. This adrenal mass is scarcely enhanced on the delayed phases.

D. An arterial enhancing nodule lies in the subcapsular portion of segment V, and retropitoneal hemorrhage is also seen (arrow).
adrenal gland, which were both embedded with extensive hemorrhage (Fig. 3A, B).

The patient had an uneventful postoperative course and was discharged. Since then, the patient has been doing well for 20 months and he has shown no evidence of tumor recurrence or metastasis.

Discussion

After the lung, the adrenal gland is the second most common site of hematogenous metastases from hepatocellular carcinoma, and this has been found in up to 8.4% of the hepatocellular carcinoma cases at autopsy (1). In another study, the lung, abdominal lymph nodes and bone were the most common sites of extrahepatic metastasis from hepatocellular carcinoma. Adrenal gland metastasis is the next most common (11%), and other unusual metastatic sites are the brain, gastrointestinal tract, spleen, seminal vesicle and bladder (3).

The causes of nontraumatic adrenal hemorrhage are categorized into five groups: stress, hemorrhage diathesis or coagulopathy, neonatal stress, underlying adrenal tumors and idiopathic disease (4). Hemorrhagic adrenal metastasis is rare, although adrenal metastases are rather common. On the other hand, primary adrenal cortical carcinoma usually contains areas of hemorrhage and necrosis to variable degrees, as seen on histologic analysis (5). Yet making the accurate differential diagno-
sis is difficult if the adrenal metastasis displays ruptured hemorrhage, unilateral involvement or necrosis. Hemorrhage from a benign lesion such as pseudocyst or myelolipoma is discernible due to the water or fat attenuation on CT scans. Pheochromocytomas characteristically appear markedly hyperintense on T2-weighted MR images and there is marked enhancement, but these tumors often contain areas of hemorrhage and also cystic components [4], so pheochromocytoma can be easily differentiated. The mechanism of spontaneous rupture by hepatocellular carcinoma and its metastasis has not been determined. It is believed that a tear in the tumor or rupture of a feeding artery causes the intratumoral pressure to increase expansively and then the hepatocellular carcinoma ruptures, and especially when the tumor has an encompassing fibrous capsule [6]. As a consequence, tearing of the adhesive surfaces or rupture of an adjacent artery occur.

Unfortunately, adrenal metastases from hepatocellular carcinoma are usually not discovered until the primary tumor becomes aggravated due to the underlying advanced liver cirrhosis, and surgery is not indicated at this phase. However, the huge adrenal mass of our patient appeared to be solitary and localized, although it was partially ruptured and the primary single hepatic tumor was small with a noncirrhotic configuration. Therefore, in such cases, surgical resection should be more strongly considered than the other treatment approaches.

An unusual enhanced pattern of the adrenal metastasis was noted during the dynamic CT scan. The CT depicted only the minimally enhanced peripheral portion and the low attenuated central necrosis of the adrenal mass. The enhanced MR imaging was not useful to differentiate adrenal metastasis from primary adenocortical carcinoma, except for the detection of the hemorrhagic foci. The poor enhancement, despite of contrast infusion, on the dynamic CT scan and on the MR images may be the result of scanty arterial vascularity, which was due to the adrenal tumor’s extensive ruptured hemorrhage.

Although, spontaneous rupture of adrenal metastasis that originates from hepatocellular carcinoma is extremity rare, we should include rupture of adrenal metastasis from hepatocellular carcinoma as part of the differential diagnosis for patients with underlying hepatocellular carcinoma.

References