1. Case report

A 45-yr-old woman was admitted for the treatment of known gynecologic diseases, including a uterine leiomyoma and an adrenal cystic mass.

Preoperative ultrasound examination incidentally detected a solid mass in the right lobe of the liver in addition to the gynecologic lesions. Unenhanced and contrast-enhanced computed tomography (CT) scans were performed to preoperatively evaluate the incidental tumor. A solid hepatic mass was detected on these CT images and was heterogeneously enhanced (Fig. 1). The maximum diameter of the lesion was 3.5 cm. However, we did not exclude the possibility of extrahepatic tumors, including tumor invasion arising from the right adrenal gland or seeding from the ovary cancer. Therefore, we made preoperative diagnoses, including cholangiocarcinoma, adrenal cortical carcinoma, and ovary cancer implant. Preoperative biochemical laboratory findings, including serum tumor markers, viral hepatitis markers, and adrenal hormones, were within normal ranges.

The patient underwent open adrenalectomy, partial hepatectomy, hysterectomy, and salpingo-oophorectomy.
The incidental adrenal mass was attached to both liver parenchyma and right adrenal gland. No peritoneal carcinomatosis was noted. Frozen section revealed no cancer cells in the incidental mass. The pathologic specimen revealed adhesion of hepatic parenchyma, right adrenal tissue, and a well-circumscribed yellowish cortical adenoma (Fig. 2). The pathologic diagnosis was confirmed to be a adrenal cortical adenoma arising from the adrenohepatic fusion tissue (Fig. 5). Microscopic examination revealed typical cortical adenoma cells with lipid-rich cytoplasm and close intermingling of adenoma cells and hepatocytes (Fig. 4). Gynecologic lesions were also removed and were confirmed to be a uterine leiomyoma and a hydrosalpinx.

When CT images were retrospectively reviewed, the adrenal tumor was hypodense compared to liver parenchyma, and the mean CT number of the lesion measured 6 Hounsfield units (HU) (range: 2–13 HU) on unenhanced images (Fig. 5). Multiplanar reformation images were created to reevaluate the origin of the adrenal tumor. The medial limb of the right adrenal gland closely abutted the liver and touched the adrenal tumor (Fig. 6), which was well correlated with the pathologic findings (Fig. 2). Indeed, we did not notice these imaging features prior to surgical excision of the tumor.

The patient had a fever at 10 d postoperatively. Postoperatively performed CT images showed a moderate amount of fluid in both hepatectomy site and pleural cavities. Passive lung collapse developed due to the bilateral pleural effusion. However, conservative treatments, including use of antibiotics and diuretics and postural drainage of sputum, were sufficient for treating the postoperative fever.
2. Discussion

Pathologically, adrenohepatic fusion is defined as fusion of the liver and right adrenal gland with closely intermingling parenchymal cells of these two organs [1]. This event does not occur so rarely but accounts for 9.9% of unselected autopsy cases [1]. It has been suggested that the mechanism of adrenohepatic fusion is that the mesenchymal tissue defect may cause retardation of capsule formation with parenchymal mixing and failure of local differentiation into fetal and later adult fat cells [2]. However, it has also been suggested that adrenohepatic fusion is an acquired event because it increases in older age [1].

Clinically, adrenohepatic fusion may become a route for cancer extension. Hepatocellular carcinoma may easily metastasize to the right adrenal gland through the adrenohepatic fusion [3] because there is no capsule between the fused parenchymal tissues. Conversely, adrenohepatic fusion is also a possible route for a malignant adrenal tumor extending to the liver parenchyma, even though it has not been reported yet. Cancer extension through an adrenohepatic fusion should be differentiated from hematogenous adrenal metastasis since patients with the former may have a better prognosis than those with the latter following adrenalectomy [3–5].

Radiologically, adrenal adenoma can be easily diagnosed on unenhanced CT images if the mass measures $\leq 10$ HU on unenhanced CT [6]. This lesion is called lipid-rich adenoma because the cytoplasm has abundant lipid [7]. Chemical shift magnetic resonance imaging (MRI) can diagnose a lipid-rich adenoma more accurately than unenhanced CT [8]. In contrast, a lipid-poor adenoma of $>10$ HU on unenhanced CT images may be more easily identified on delayed contrast-enhanced CT than chemical shift MRI [9]. Woo et al reported that adrenal cortical adenoma developing from adrenohepatic fusion tissue in an oncologic patient may be mistaken for a metastatic lesion [10]. In their report, however, there was no diagnostic clue to indicate an adrenal cortical adenoma on the preoperative CT images. In a retrospective review of CT imaging features, a diagnosis of adrenal adenoma should have been made easily because it measured $\leq 10$ HU on unenhanced CT images. None of the solid hepatic or adrenal tumors except tumors containing gross fatty tissue are as hypodense as adrenal adenoma on unenhanced CT images. Additionally, three-dimensional reconstruction of CT images obtained from multi-detector-row CT examination can depict the right adrenal gland touching the mass arising from adrenohepatic fusion tissue [10].

In conclusion, an adrenal adenoma arising from the adrenohepatic fusion tissue might be precisely diagnosed if the lesion measures $\leq 10$ HU on unenhanced CT and is continuous with a right adrenal gland on multiplanar reformation images. Therefore, these imaging features may help avoid unnecessary surgical procedures.

Conflicts of interest: The authors have nothing to disclose.

EU-ACME question

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Question:
What is the most probable diagnosis of an adrenal solid mass measuring <10 Hounsfield units (HU) on unenhanced computed tomography (CT) images?

A. Metastasis  
B. Cortical carcinoma  
C. Pheochromocytoma  
D. Cortical adenoma

References