Clinical Case Report

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Intrahepatic adrenocortical adenoma arising from adrenohepatic fusion mimicking hepatic malignancy

Two case reports

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Abstract

Rationale: Intrahepatic adrenocortical adenoma (IAA) arising from adrenohepatic fusion (AHF) is rare and its imaging findings are not well established. Moreover, it is easily misdiagnosed as malignant hepatic tumor in patients at risk of malignancy. Its key finding is the connection between the tumor and adrenal gland. When IAA from AHF is suspected, biopsy should be considered to avoid unnecessary surgery. Herein, we report 2 cases of IAA from AHF.

Patient concerns: A 59-year-old woman was admitted due to a 1.5-cm hypoechoic nodule in the right hepatic lobe detected on ultrasound for hepatocellular carcinoma (HCC) surveillance due to chronic hepatitis B. Contrast-enhanced computed tomography (CT) and gadoxetic acid-enhanced magnetic resonance imaging (MRI) were performed to evaluate the hepatic mass. Another 75-year-old woman was admitted due to rectal adenocarcinoma detected on colonoscopy. Contrast-enhanced CT depicted a 2.5-cm mass in the right hepatic lobe.

Diagnosis: In case 1, CT and MRI showed a 1.5-cm subcapsular mass in the right hepatic lobe with typical findings of HCC in a patient with chronic hepatitis B. The mass was confirmed as IAA from AHF after the laparoscopic surgery. In case 2, CT showed advanced rectal malignancy and a 2.5-cm poorly enhancing mass in the right hepatic lobe. The tentative diagnosis was hepatic metastasis. However, based on the connection between the tumor and adrenal gland during preoperative review, the presumed diagnosis was changed to IAA from AHF, which was confirmed on biopsy.

Interventions: The hepatic mass connected with the right adrenal gland was laparoscopically resected in case 1. Laparoscopic lower anterior resection for rectal malignancy and percutaneous biopsy for the hepatic mass were performed in case 2.

Outcomes: The first patient had an uneventful recovery, without recurrence on the 3-year follow-up CT. The second patient had an uneventful postoperative course and has been alive for 12 months postoperatively without pathologically proven IAA changes on follow-up CT.

Lessons: When hepatic mass is found adjacent to the right adrenal gland on imaging, the connection between the tumor and adrenal gland should be investigated. When IAA arising from AHF is suspected, biopsy should be considered to avoid unnecessary surgery.

Abbreviations: AHF = adrenohepatic fusion, CT = computed tomography, HCC = hepatocellular carcinoma, IAA = intrahepatic adrenocortical adenoma, MRI = magnetic resonance imaging.

Keywords: adrenohepatic fusion, biopsy, computed tomography, intrahepatic adrenocortical adenoma, magnetic resonance imaging

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1. Introduction

Intrahepatic adrenocortical adenoma (IAA) arising from adrenohepatic fusion (AHF) is rare; however, it could mimic malignant hepatic tumor in patients at risk of malignancy. In patients at risk of primary malignancy, IAA arising from AHF is frequently misdiagnosed as primary hepatic malignancy such as hepatocellular carcinoma (HCC) because of its rarity and the lack of well-established imaging findings.[1–3] In patients at risk of metastatic hepatic tumor, unnecessary surgery or chemotherapy could be performed or patients could lose the chance of curative surgery due to misdiagnosed metastasis of IAA arising from AHF. We experienced 2 cases in which the connection between the tumor and right adrenal gland could be a key imaging finding and percutaneous core needle biopsy was helpful for confirmative diagnosis. Therefore, we report 2 cases with review of the previous literature.

2. Case report

2.1. Case 1

A 59-year-old woman was admitted for the evaluation of a 1.5-cm hypoechoic subcapsular nodule in the right hepatic lobe detected during the ultrasound screening examination for HCC surveillance due to chronic hepatitis B. Physical examination revealed no characteristic features. Laboratory tests upon admission showed her serum was positive for hepatitis B core antigen and hepatitis B surface antigen, but negative for hepatitis B surface and hepatitis C virus antibodies. Other laboratory values including aspartate aminotransferase, alanine aminotransferase, total bilirubin, alkaline phosphatase, and alpha-fetoprotein were unremarkable.

She underwent dynamic contrast-enhanced computed tomography (CT) to evaluate the right hepatic mass. CT images showed a 1.5-cm subcapsular round mass with hyper-arterial enhancement and delayed washout in the posterior segment at the right hepatic lobe, which was suspected as HCC (Fig. 1A). In order to distinguish the tumor between the HCC and other hepatic tumors, gadoxetic acid-enhanced liver magnetic resonance imaging (MRI) was performed, which also depicted a 1.5-cm well-demarcated round subcapsular mass with typical imaging findings of HCC such as hyper-arterial enhancement, delayed washout, and hypointensity on hepatobiliary phase on gadoxetic acid-enhanced MR images (Fig. 1B and C). The tumor was diagnosed as a HCC based on the typical findings seen on dynamic contrast-enhanced CT and gadoxetic acid-enhanced MRI and the history of chronic hepatitis B in accordance with the American Association for the Study of Liver Diseases (AASLD)

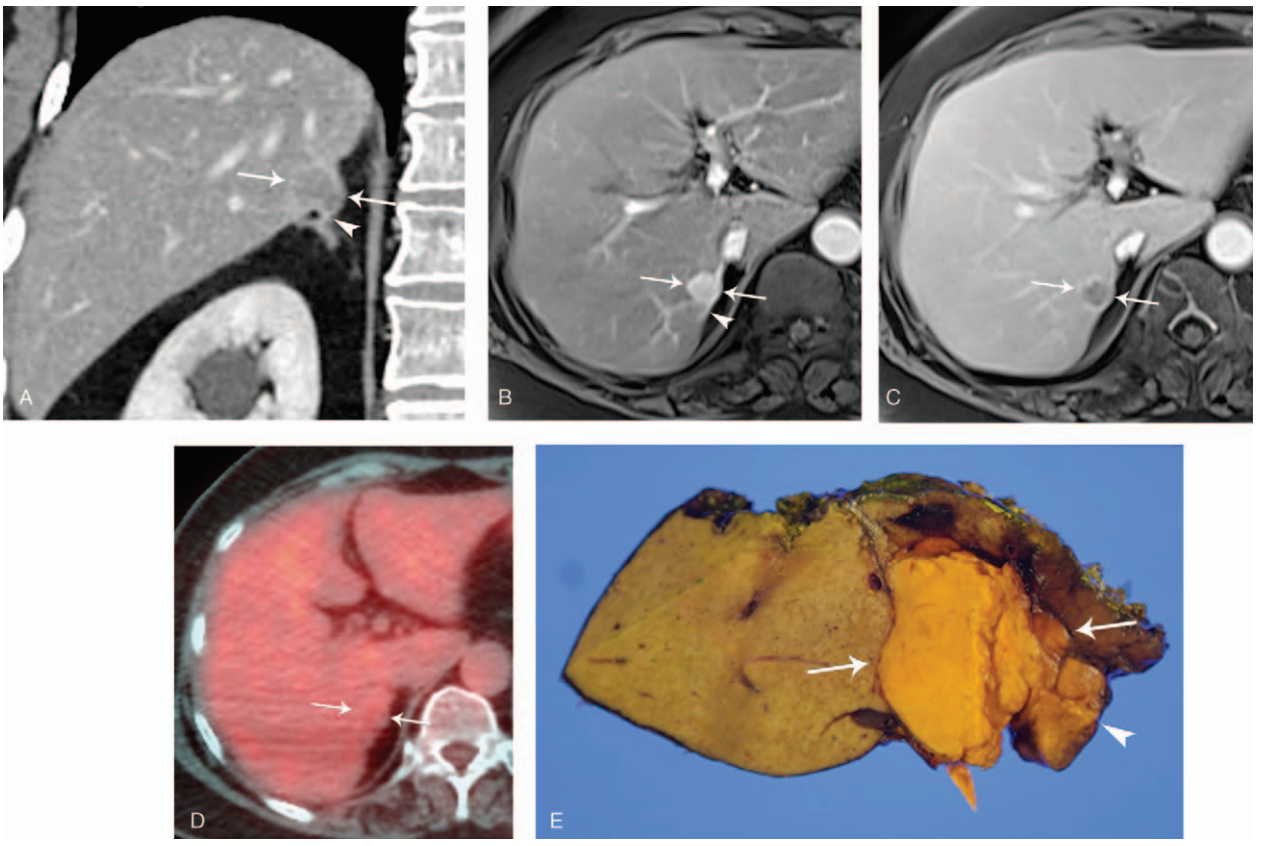

Figure 1. A 1.5-cm intrahepatic adrenocortical adenoma in a 59-year-old woman that was pathologically confirmed through the right posterior sectionectomy. (A) Contrast-enhanced coronal portal phase CT image clearly shows the connection (arrowhead) between the tumor (arrows) and lateral limb of right adrenal gland, which implies the tumor arising from adrenohepatic fusion. (B and C) Gadoxetic acid-enhanced MR images demonstrate hyper-arterial enhancement and delayed washout of a round subcapsular tumor (arrows), such as the typical enhancement pattern of hepatocellular carcinoma, in the right hepatic lobe. Note the arterial enhancing linear lateral limb (arrowhead) in the right adrenal gland, which is continuous with the tumor on B. (D) ^{18}F-FDG-PET/CT image depicts the ^{18}F-FDG uptake of the tumor (arrows) similar to the physiologic uptake of the liver parenchyma, which suggests low likelihood of malignancy. (E) Photograph demonstrates a 1.5-cm yellowish subcapsular tumor (arrows) of the resected liver, which is connected to the right adrenal gland (arrowhead).

guideline.[4] According to AASLD guideline, laparoscopic right posterior sectionectomy was decided by a multidisciplinary tumor board consisting of hepatologists, hepatic surgeons, oncologists, and abdominal radiologists at our hospital. Although the tumor was not depicted as a lesion with hot uptake on ^{18}F-FDG-PET/CT image (Fig. 1D), presumed diagnosis was HCC because the patient had chronic hepatitis B, which is high risk for HCC.

In the operative field, the surgeon identified a 1.5-cm well-circumscribed, capsulated white-to-yellow hepatic mass that was connected with the right adrenal gland. The cut surface of the tumor was yellowish and showed signs of fat component (Fig. 1E). IAA arising from AHF was pathologically confirmed by laparoscopic surgical resection and microscopic findings. The patient had an uneventful recovery and was discharged on postoperative day 7. No tumor recurrence in the liver was noted on the 3-year follow-up CT.

2.2. Case 2

A 75-year-old woman was admitted due to a rectal nonmucinous adenocarcinoma detected on colonoscopy during a medical screening program. Her history was otherwise noncontributory. A physical examination revealed no characteristic features. Laboratory tests upon admission were unremarkable. She underwent abdominal dynamic contrast-enhanced CT for staging of the rectal malignancy, which showed advanced upper rectal malignancy with perirectal invasion and regional lymphadenopathies. A 2.5-cm lobulated well-defined poorly enhancing subcapsular mass in the posterior segment of the right hepatic lobe was also detected on CT images (Fig. 2A and B). The hepatic tumor was tentatively diagnosed as a solitary hepatic metastasis from rectal malignancy at the time of the staging. However, a connection between the hepatic tumor and right adrenal gland was found on the coronal CT image during careful review by a multidisciplinary tumor board for appropriate treatment plan (Fig. 2C). This imaging finding is considered as a key factor to diagnose IAA arising from AHF, although its incidence is rare and its imaging findings are not well established. Thus, percutaneous ultrasound-guided core needle biopsy was performed to distinguish between hepatic metastasis and IAA arising from AHF (Fig. 2D). The results of immunohistochemical staining for the obtained tumor tissue were negative for hepatocyte-specific antigen and positive for melan-A and alpha-inhibin, and the tumor was pathologically confirmed as IAA arising from AHF (Fig. 2E).

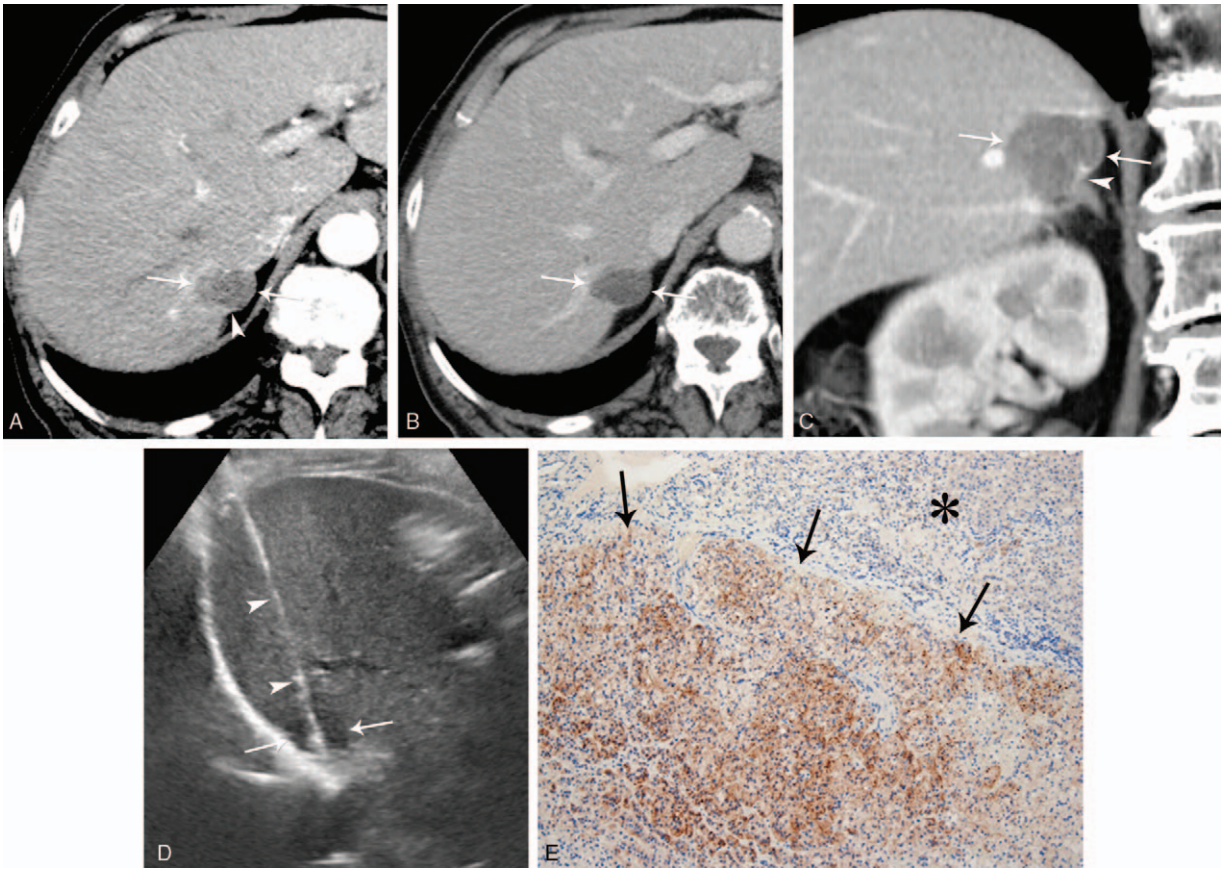

Figure 2. A 2.5-cm intrahepatic adrenocortical adenoma in a 75-year-old woman that was pathologically confirmed using ultrasound-guided percutaneous core needle biopsy. (A–C) Contrast-enhanced CT images demonstrate a poorly enhancing mass (arrows) in the right hepatic lobe, which was connected with the lateral limb of the right adrenal gland (arrowhead). (D) Oblique transverse intercostal ultrasound image shows a hypoechoic mass (arrows) in the right hepatic lobe. Ultrasound-guided percutaneous core needle biopsy with an 18-guage needle (arrowheads) using an intercostal approach was performed while the patient was in the left posterior oblique position. No complication occurred after biopsy. (E) Microscopic image of melan-A immunohistochemical staining (specimen of core needle biopsy, ×100) demonstrates that adrenocortical adenoma cells (lower two-third, arrows) show positivity and hepatocytes (upper one-third, asterisk) demonstrate negativity.

She underwent laparoscopic lower anterior resection with loop ileostomy for rectal malignancy and adjuvant chemotherapy using oxaliplatin and 5-fluorouracil. She had an uneventful postoperative course and has been alive for 12 months postoperatively. The pathologically proven adrenocortical adenoma arising from AHF showed no changes on follow-up CT images.

3. Methods

The patients provided signed, informed consent for publication of the cases, and the study was approved by the Institutional Review Board of Chonnam National University Hwasun Hospital (CNUHH-2019-068).

4. Discussion

AHF is known as the union between the right adrenal gland and liver by intermingling its parenchyma. Honma reported that AHF was found in 63 of 636 unselected autopsies (9.9%), suggesting that this was a rather common anatomic condition. Moreover, older age groups had a higher incidence of AHF.[5] Adrenocortical adenoma from AHF is now more commonly detected in the clinical practice than what was known because imaging techniques such as CT and MR have been developed and increasingly used. However, adrenocortical adenomas in the liver have been rarely encountered. Most of the reported cases were adrenal rest tumors, that is, adrenal cortical tumors originated from the accessory adrenal tissue in the liver with intact adrenal glands in their normal position.[6–8] Because IAA arising from AHF is rare, the clinical presentation and imaging findings of the tumor have been rarely described in the literature. Most IAAs have been detected incidentally during imaging work-up, especially in patients with known malignancy.

Adrenocortical adenoma is highly suggested when the HU measurement is less than 10 on unenhanced CT.[9,10] This characteristic imaging feature is based on the presence of intralesional fat. Typical adrenocortical adenoma was demonstrated as early enhancement on the arterial phase and delayed washout pattern on far delayed phase images. For these reasons, the IAA arising from AHF can mimic HCC on cross-sectional imaging.[11] In previous reports, most adenoma cases from AHF were preoperatively misdiagnosed as primary or metastatic hepatic tumors, especially HCC.[1–3,12,13] As mentioned, imaging findings of IAA from AHF tend to mimic those of HCC, making its differentiation very difficult in patients highly at risk for HCC. That is why most of the reported cases and our first case were misdiagnosed as HCC. In case 1, the subcapsular hepatic mass was preoperatively misdiagnosed as HCC due to chronic hepatitis B history and typical enhancement pattern of HCC on CT and MRI. However, low ^{18}F-FDG uptake within the tumor similar to the liver parenchyma and low value of alpha-fetoprotein suggest low likelihood of HCC. Moreover, because the imaging finding of IAA from AHF has not been well established, the connection between the tumor and right adrenal gland was not considered, which implies the tumor arising from AHF. In case 2, a subcapsular hepatic mass was initially diagnosed as a single hepatic metastasis due to the preexisting rectal malignancy and poorly enhancing hepatic mass on CT. However, the connection between the tumor and right adrenal gland attracted our attention in the preoperative imaging reviews. Thus, we considered the possibility of IAA from AHF and performed US-guided core needle biopsy to confirm our diagnosis. Similar with our 2 cases, a hepatic mass is demonstrated adjacent to the right adrenal gland on the cross-sectional imaging, the possible connection between the tumor and adrenal gland should be carefully investigated using multiplanar reconstruction cross-sectional images, which could be a key finding for correct diagnosis.

Accurate diagnosis of IAA arising from AHF is important for pathologic confirmation as well as for preoperative diagnosis. Pathologically differentiating IAA in AHF from HCC is difficult without clinical suspicion of IAA, due to their similar histological findings.[3,7] Immunohistochemical staining using hepatocyte-specific antigen, melan-A, or alpha-inhibin is useful to differentiate IAA from HCC. Immunohistochemical staining of IAA is generally negative for hepatocyte-specific antigen and positive for melan-A or alpha-inhibin and that of HCC is contrary to the results of IAA.[7]

Although the imaging findings of IAA in AHF are similar to those of primary or metastatic hepatic tumors, the connection between the tumor and right adrenal gland could be a key finding for accurate diagnosis. Moreover, low ^{18}F-FDG uptake on PET/CT could help diagnose IAA arising from AHF. When IAA arising from AHF is suspected on imaging, percutaneous core needle biopsy should be considered. In addition, the obtained specimen should be evaluated for the possibility of IAA arising from AHF in order to avoid unnecessary surgery.

Author contributions

Conceptualization: Jin Woong Kim, Young Hoe Hur.
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Supervision: Jin Woong Kim, Jun-Hee Park, Young Hoe Hur.
Writing – original draft: Yong Soo Cho.
Writing – review & editing: Jin Woong Kim, Hyun Ju Seon, Jun-Hee Park, Hyung Joong Kim, Young Hoe Hur.

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