



Extrauterine Adenomyoma of the Liver Mimicking a Hepatic Adenoma: A Case Report

간선종으로 오인된 간에 발생한 자궁외 선근종: 증례 보고

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Extrauterine adenomyoma of the liver is extremely rare. Only a few cases have been reported, and these reports have focused mainly on histopathology. Herein, we report the specific imaging findings of extrauterine adenomyoma of the liver in a 43-year-old woman with epigastric pain, which was initially diagnosed as a hepatic adenoma. CT and MRI revealed a solid and cystic mass with hemorrhagic foci and weak persistent enhancement, located in the subcapsular region of the right hepatic lobe.

Index terms Adenomyoma; Liver; Adenoma, Liver Cell; Computed Tomography, X-Ray; Magnetic Resonance Imaging

INTRODUCTION

Extrauterine adenomyomas are defined as circumscribed tumor-like masses composed of smooth muscle tissue, endometrioid glands, and stroma and they are similar to their more common uterine counterparts in most respects (1). They are extremely rare, especially those originating in the liver. To date only 3 cases of hepatic extrauterine adenomyoma have been reported, and those reports are focused on histopathology (2).

Herein, we report for the first time the specific radiologic findings of extrauterine adenomyoma of the liver in a 43-year-old woman, which was initially diagnosed as a hepatic adenoma.

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CASE REPORT

A 43-year-old woman presented at the digestive internal medicine department with a 7-day history of epigastric pain. A general physical examination and a systemic examination were unremarkable. She had undergone total laparoscopic hysterectomy for uterine leiomyoma 4 years prior. Otherwise, she had no underlying medical diseases. Initial laboratory evaluations including tumor marker (alpha-fetoprotein) and liver function tests were within normal ranges, and serological tests for hepatitis B surface antigen and anti-hepatitis C virus antibodies were negative.

Upper gastrointestinal endoscopy was performed to investigate her epigastric pain, and there were no abnormal findings. Contrast-enhanced CT scanning was performed to further investigate her symptoms. It revealed a $3 \times 2 \times 2$ cm well defined bilobed mass in the subcapsular area of segment VI of the liver. The medial lobe of the mass exhibited low density without enhancement, and the lateral lobe exhibited an enhanced solid portion with peripheral low-density dots. The lateral lobe was shaped like a lotus root. The enhancement pattern was subtle enhancement in the arterial phase and persistent weak enhancement in the portal and delayed phases without washout (Fig. 1A). The CT characteristics of the lesion did not facilitate a definitive diagnosis, so we recommended MRI.

On MRI, the medial lobe and the periphery of the lateral lobe that exhibited low density on CT were depicted as cystic portions that exhibited high signal intensity on T2-weighted images. There was focal high signal intensity with fluid-fluid level within the medial cystic portion on in-phase and out-of-phase T1-weighted images, suggesting a hemorrhagic component. The solid portion of the lateral lobe of the mass exhibited subtle low signal intensity on T1-weighted images and slightly high signal intensity on T2-weighted images. Gadolinium-enhanced fat-suppressed T1-weighted images depicted heterogenous weak arterial enhancement and persistent enhancement (Fig. 1B).

The patient underwent wedge resection of hepatic segment VI. Intraoperatively a mass of approximately $3 \times 2 \times 2$ cm in size was located in the subcapsular region in segment VI of the liver. A gross specimen photograph is shown in Fig. 1C. Wedge resection of the liver revealed a bilobed mass located subcapsularly. The mass was well defined, whitish, and solid with multiple cysts filled with dense brown fluid. Microscopically, the mass was composed of thick muscular tissue lined with benign endometrial gland and stroma that resembled uterine endometrium. The endometrioid glands were positive for estrogen receptor and progesterone receptor, and the endometrioid stromas were positive for CD10. The smooth muscle component was positive for smooth muscle actin (Fig. 1D).

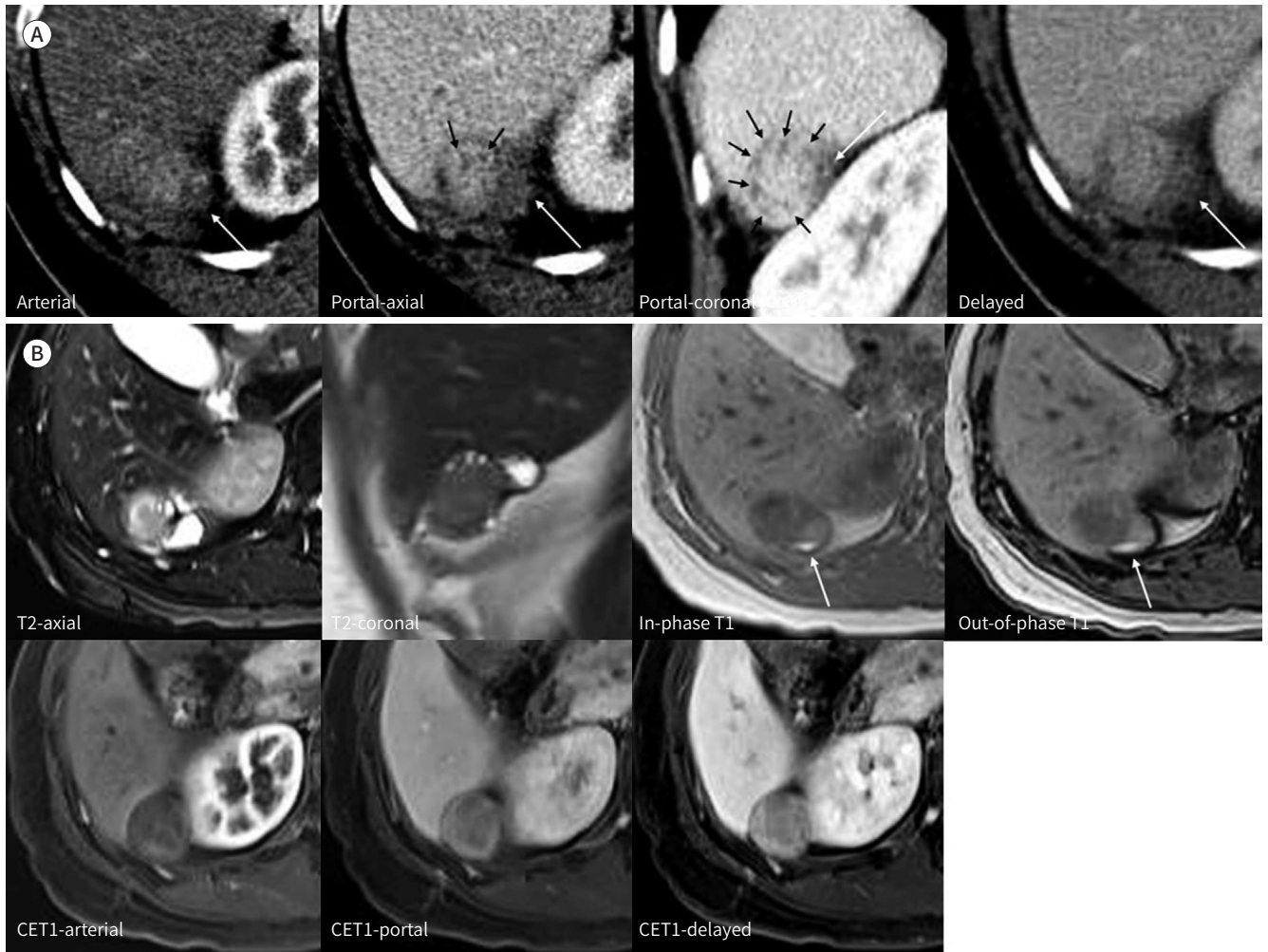
DISCUSSION

Adenomyomas are benign tumors consisting of benign endometrial glands, endometrioid stroma, and smooth muscle tissue, and they typically originate within the uterus (1). Occasionally they are located outside the uterus, and such tumors are termed extrauterine adenomyomas. The majority of such cases originate from the ovary and are located in the pelvis, and extrapelvic locations such as the liver or abdominal wall are extremely rare. The first

Fig. 1. Extrauterine adenomyoma of the liver in a 43-year-old woman.

A. Axial contrast-enhanced CT reveals a $3 \times 2 \times 2$ cm bilobed mass in the subcapsular area of segment VI of the liver. The medial lobe of the mass exhibits low-density without enhancement (white arrows), and the lateral lobe exhibits an enhanced solid portion with peripheral low-density dots (black arrows). The enhancement pattern is subtle in arterial phase, with persistent enhancement in the portal and delayed phases without washout.

B. Axial and coronal T2-weighted images exhibit high-signal intensities in the medial lobe and periphery of the lateral lobe of the bilobed mass, indicating a cystic component. There is focal high-signal intensity within the medial cystic portion on the in-phase and out-of-phase axial T1-weighted images, suggesting a hemorrhagic component (arrows). The solid portion of the lateral lobe of the mass exhibits slight high-signal intensity on T2-weighted images, and a subtle low-signal intensity on T1-weighted images. It also exhibits mild heterogeneous enhancement in the arterial phase and weak persistent enhancement in the portal and delayed phases.



case of extrauterine adenomyoma was reported by Paul et al. (2) and Cozzutto in 1981 (3) reported that there had been 37 cases of extrauterine adenomyoma eligible for their review since then. Of these 37 cases, 21 were pelvic, 8 were extrapelvic, and 8 cases involved multiple sites. In their report there were only 3 cases of extrauterine adenomyoma involving the liver, and these reports focused on histopathology (1, 4, 5).

The above-mentioned previously reported 3 cases were only evaluated via abdominal CT, and all of the masses were typically located in the subcapsular region of the posterior right hepatic lobe. All had a cystic portion but there were no definite intralesional hyperattenuating hemorrhages on CT. However, there were hemorrhagic components on gross and patho-

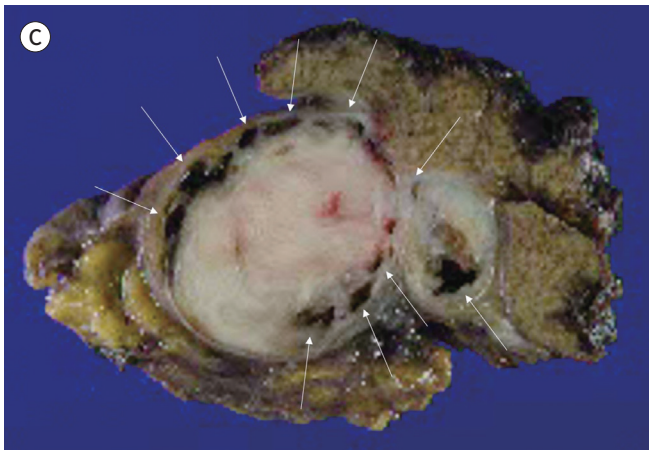
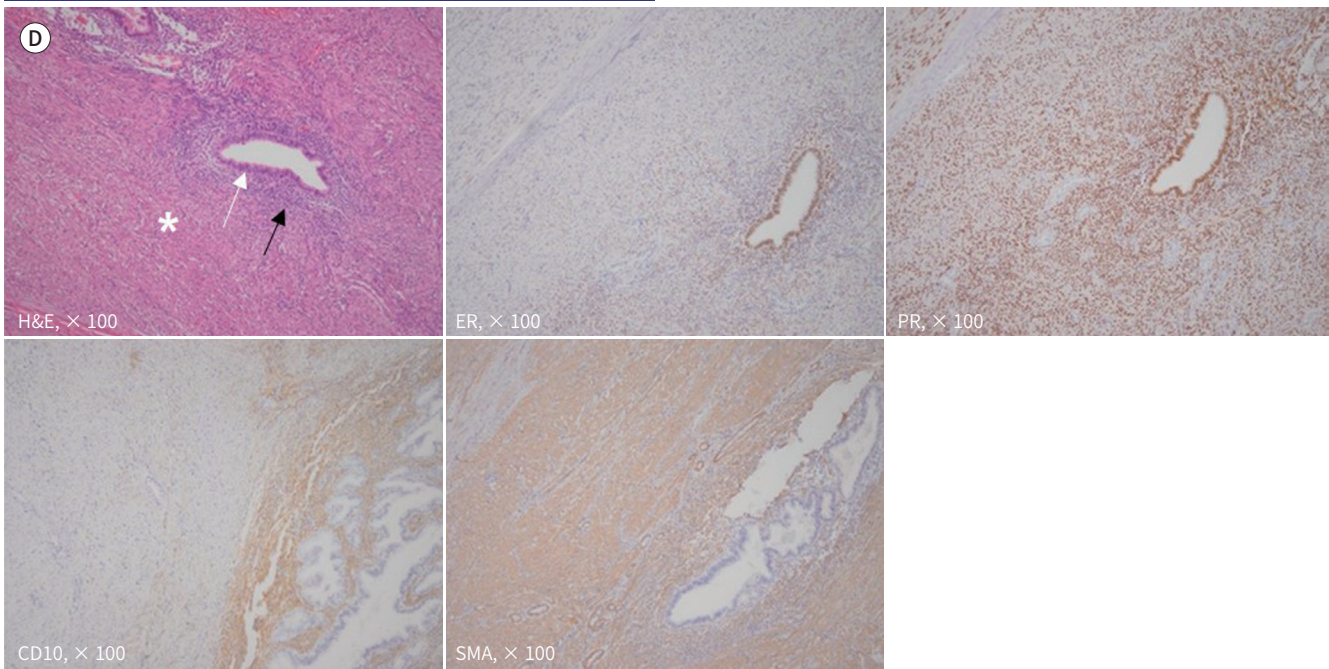


Fig. 1. Extrauterine adenomyoma of the liver in a 43-year-old woman. **C.** Photograph depicting the gross appearance of the mass shows well defined, bilobed, whitish, and solid mass with multiple cysts filled with dense brown fluid (arrows).

D. Microscopically, the mass is composed of thick muscular tissue (asterisk) lined with benign endometrial gland (white arrow) and stroma (black arrow) that resembles uterine endometrium (H&E). ER and PR immunohistochemical stains are suggestive of endometrioid glands. The endometrioid stromas are positive for CD10. The smooth muscle component is positive for SMA.

ER = estrogen receptor, H&E = hematoxylin and eosin stain, PR = progesterone receptor, SMA = smooth muscle actin



logic findings. Two exhibited heterogenous arterial enhancement and the remaining 1 was not evaluated via contrast-enhanced CT. In the present case the mass was also located in the subcapsular area of the posterior right hepatic lobe, and it exhibited subtle arterial enhancement and weak persistent enhancement in the portal and delayed phases. It had cystic portions with hemorrhagic foci. We surmise that these locational and morphologic findings are specific imaging features of extrauterine adenomyoma of the liver.

Morphologic imaging finding of uterine adenomyomas and extrauterine adenomyomas including liver and other sites is similar in that they are solid and cystic masses with some internal hemorrhagic foci and show heterogenous enhancement (2, 6).

Because the most common benign hepatic mass with hemorrhage is adenoma, of which inflammatory subtype can exhibit enhancement during the arterial phase that persists in the portal and delayed phases (7) we initially mistook the present case for hepatic adenoma. However, hepatic extrauterine adenomyoma is likely to contain a cystic and hemorrhagic component anywhere within it, and hepatic adenoma is prone to central necrosis and hem-

orrhage because the vascular supply is limited to the surface of the tumor. Furthermore, hepatic adenoma can have a fat component and pseudocapsule in as many as 30% of cases (7, 8). Otherwise, hepatic endometrioma, abscess, hemangioma, hepatocellular carcinoma (HCC), hemorrhagic metastasis can also show solid and cystic mass with intralesional hemorrhage. However, unlike hepatic adenomyoma having a dominant solid smooth muscle component with scattered endometrial glands which show cystic and hemorrhagic foci, hepatic endometrioma has less or no solid component of smooth muscle and usually shows predominant cystic lesion with intralesional hemorrhage (9). Hepatic abscess typically demonstrates rim enhancement with perilesional edema and hemangioma exhibits centripetal enhancement pattern. HCC also reveals typical enhancement pattern that arterial enhancement and delayed washout. Hemorrhagic metastasis can show one or more liver lesions with primary tumor elsewhere (7).

The mechanism of hepatic extrauterine adenomyoma is still uncertain. However, various theories have been proposed to explain the pathogenesis of extrauterine adenomyoma. In our report, we are discussing 2 of the major theories. The first theory is the implantation theory. All patients of 3 previously reported hepatic adenomyoma cases and our case had undergone laparoscopic surgeries such as myomectomy or hysterectomy for uterine leiomyoma and hysterectomy with salpingectomy for adenomyosis, leiomyoma and endometriosis. During surgery, peritoneal fluid may have viable endometrial cells and smooth muscle cells from cut uterus. And these could experimentally be implanted and grown in a peritoneum or peritoneal cavity. The second theory is the sub-coelomic mesenchyme transformation theory that extrauterine adenomyoma originates from the metaplasia of the layer that lies underneath the mesothelium of the abdominal peritoneum. The cases of hepatic extrauterine adenomyomas including our case were located in the subcapsular region of the liver, which was partially covered by the hepatic peritoneum (1, 9, 10).

Clinically, patients with hepatic extrauterine adenomyoma can present with abdominal pain or back pain (1, 4, 5). It may be that pain is caused when hemorrhage occurs in the mass.

In summary, a definitive diagnosis of extrauterine adenomyoma is only established post-operatively after confirmation via histopathology. However, extrauterine adenomyoma of the liver exhibits specific radiologic features such as a subcapsular location in the posterior right hepatic lobe, a weak persistent enhancement pattern, and a cystic portion with internal hemorrhage. Therefore when this kind of benign hemorrhagic mass of the liver is encountered, extrauterine adenomyoma should be considered.

Author Contributions

Conceptualization, W.Y.J., W.J.Y., B.J.; data curation, W.Y.J., H.M.E.; formal analysis, W.Y.J.; funding acquisition, W.Y.J., W.J.Y.; investigation, W.Y.J.; methodology, W.Y.J., B.J.; project administration, W.Y.J., W.J.Y.; resources, W.Y.J.; software, W.Y.J.; supervision, W.Y.J., W.J.Y.; validation, W.Y.J.; visualization, W.Y.J., H.M.E.; writing—original draft, W.Y.J.; and Writing—review & editing, W.Y.J., W.J.Y., B.J.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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간선종으로 오인된 간에 발생한 자궁외 선근종: 증례 보고

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간에 발생하는 자궁외 선근종은 매우 드문 종양이다. 이제껏 몇 가지의 사례만이 문헌에 보고되었고, 이것들은 조직병리학 소견에 초점을 둔 보고들이었다. 우리는 처음에 간선종으로 오인되었던, 상복부 통증을 주소로 내원한 43세 여성에서의 간에 발생한 자궁외 선근종의 특징적인 영상 소견을 보고하고자 한다. 이것은 전산화단층촬영과 자기공명영상에서 국소적 출혈을 동반한 고형 및 낭성 종괴로 보이며 간우엽 피막하에 위치하였고, 지속적인 약한 조영 증강을 보였다.

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