Splenic hemangiomas with different imaging findings: two laparoscopically resected incidentalomas

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Abstract

Although splenic tumors are relatively rare, we sometimes encounter incidentalomas in the spleen. In such cases, we must plan treatment based on imaging studies. Herein, we report two cases of splenic hemangioma with different imaging findings, which were resected laparoscopically.

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Key Clinical Message (<50 words)

Although splenic tumors are relatively rare, we sometimes encounter incidentalomas. We must plan treatment based on imaging studies. In some cases, we might have to consider detailed imaging study and/or tumor biopsy.

Introduction

Splenic tumors are rare. They include benign primary tumors, such as lymphangioma and hemangioma, and malignant primary tumors, such as malignant lymphoma and angioblastoma. Also, solid cancers, such as gastric, colon, and ovarian cancer sometimes metastasize to the spleen. In 2013, Heller suggested a diagnostic and follow-up strategy for splenic tumors as shown in Figure 1 (1); however, it is currently not easy to differentiate between the various histological types of splenic tumors and choose an appropriate treatment plan. Herein, we report two cases of splenic hemangioma with different imaging findings. The tumors mimicked malignant tumors, which led us to perform laparoscopic splenectomy.

Case history/ Examination

Case 1

A female in her 50s presented to our internal medicine department to undergo a work-up after multiple splenic tumors had been detected by ultrasound during a health check-up. She did not have a fever or abdominal pain. Her medical history was non-specific, and she had not previously suffered from a panic disorder or appendicitis in childhood. She had been taking clotiazepam for a while. She was not in acute distress, and her vital signs were non-specific. On physical examination, her spleen was not palpable, and she did not have lymphadenopathy.

Case 2

A female in her 80s presented to our surgical department to undergo follow-up examinations for ascending colon cancer, which had been resected one year ago. She had a mild discomfort in the left upper quadrant. She did not have a fever. She had undergone laparoscopic right hemicolectomy for advanced ascending colon cancer (pT4a N1 M0, Stage IIIa, according to the Japanese Classification of Colorectal, Appendiceal, and Anal Carcinoma, Nineth English Edition) one year ago. She had not received adjuvant chemotherapy according to her wishes. Otherwise, her medical history was unremarkable and did not include hypertension or hyperlipidemia. On physical examination, her spleen was not palpable, and she did not have lymphadenopathy.

Differential diagnosis, investigations, and treatment

Case 1

A contrast-enhanced computed tomography (CT) scan (Fig. 2a, b) showed innumerable splenic tumors, which were homogeneous, but poorly demarcated. The maximum diameter of the largest tumor was 3 cm. The tumors were not enhanced. The attending physician decided to perform a follow-up CT scan 6 months later.

The patient was referred to our surgical department to undergo a more detailed consultation at that time. A laboratory workup demonstrated a normal complete blood count, an elevated lactate dehydrogenase level (404 IU/L), and a slightly elevated cancer antigen 19-9 (CA19-9) level (58.1 U/mL), but a normal carcinoembryonic antigen (CEA) level (2.3 ng/mL). A follow-up contrast-enhanced CT scan showed slight enlargement of the tumors (maximum diameter: 3.5 cm) without any findings that were indicative of diseases affecting other organs, such as solid cancer (Fig. 2c, d). No specific findings were noted during upper or lower endoscopy. We could not rule out a malignant tumor because of the following: 1. The tumors exhibited poor demarcation. 2. The tumors were slightly enlarged. 3. The patient's CA19-9 level was slightly elevated. Although Heller's strategy recommends positron emission tomography/CT or a biopsy as a next step, since the tumors were innumerable and small, we performed laparoscopic splenectomy as a "diagnostic treatment" (Fig. 3).

Case 2

A contrast-enhanced CT scan performed 6 months after the colon surgery showed a solitary splenic tumor (maximum diameter: 2.0 cm) without any other organ metastasis or dissemination. The tumor was homogenous and well-demarcated (Fig. 4a, b). Twelve months later, a follow-up contrast-enhanced CT scan showed marked enlargement of the splenic tumor (maximum diameter: 2.5 cm), without any other organ metastasis or dissemination (Fig. 4c, d). A laboratory workup demonstrated normal tumor marker levels (CEA: 5.2 ng/mL, CA19-9: <0.5 U/mL). We suspected metastatic cancer due to the rapid progression of the disease. Since the patient did not have any other organ metastasis, we performed laparoscopic splenectomy as a "diagnostic treatment" (Fig. 5).

Outcome and follow up

Both operations were performed without any postoperative complications. Histopathologically, both tumors were diagnosed as hemangiomas. Case 1: Multiple nodules were found in the spleen, which microscopically consisted of proliferating blood vessels without epithelial abnormalities (Fig. 6). Case 2: A cystic nodule

was found in the spleen, which microscopically consisted of proliferating blood vessels without epithelial abnormalities (Fig. 7, 8). We followed the patients for 3 months after surgery.

Discussion

Splenic tumors are relatively rare. In fact, Bostick WL encountered 10 cases among 80,527 people (2). There are various histological types, including benign primary tumors, such as lymphangioma, hamartoma, and hemangioma; malignant primary tumors, such as malignant lymphoma and angioblastoma; and metastatic tumors (3-5). The most common type of primary benign splenic tumor is hemangioma, almost all of which occur as solitary lesions (6). However, there have been a few reports about cases involving multiple splenic tumors (7) (8). On the other hand, malignant primary splenic tumors, such as angioblastoma, are very rare. However, their prognosis is quite poor (6). The clinical presentation of splenic tumors is highly variable (9); therefore, we must learn how to differentiate among them. Although Heller suggested a diagnostic and follow-up strategy for splenic tumors, which is currently used worldwide (1), challenging cases are sometimes encountered.

We experienced two cases of splenic hemangioma with different clinical presentations and imaging findings.

In case 1, we suspected angiosarcoma because of the following findings: 1. The tumors enlarged. 2. The tumors were heterogeneous and poorly demarcated. (10) Although we initially considered hemangioma, large splenic tumors sometimes mimic angioblastoma (11). In cases of splenic angiosarcoma, surgery is the only curative treatment, and hence, the chance to resect such tumors must not be missed. This was the reason why we performed splenectomy. It might have been better to have conducted a magnetic resonance imaging (MRI) scan. S.Y. Choi reported that contrast-enhanced dynamic and diffusion-weighted MRI are useful for distinguishing between benign and malignant splenic tumors (12).

In case 2, we suspected a metastatic tumor derived from adenocarcinoma of the ascending colon because of both the patient's medical history and the rapid enlargement of the tumor. Solitary splenic metastases are rare, but they can occur in some cases (13). It may have been better to perform a biopsy of the tumor. Although in Japan splenic biopsies are hardly performed due to fears of causing intraabdominal bleeding or tumor dissemination, they are reported to be a safe and effective way of distinguishing among splenic tumors (14) (15) (16) (17) (18). Heller recommended that biopsies should be performed as a second diagnostic step for splenic tumors that are suspected of malignancy (1). On the other hand, Cho reported a case in which severe complications occurred after a biopsy of a splenic tumor (19). All things considered, biopsying splenic tumors is quite effective; however, with recognition of the potential complications, clinicians should take sufficient precautions such as antibiotic therapy, and preparation for an urgent surgery (splenectomy in some cases).

Laparoscopic splenectomy for hemangioma was first performed by Hodge in 1895 (8). Due to a fear of bleeding, the hand-assisted method is usually selected in the cases involving giant splenomegaly. For splenic tumors, laparoscopic splenectomy is superior to classical open surgery because the laparoscope makes it possible to see the whole abdomen from various angles. This was particularly useful in our case 2, as we initially suspected that the tumor was a metastatic lesion, and hence, it was important to rule out other types of metastases, such as dissemination.

In summary, there are a wide variety of splenic tumors. The Heller's diagnostic pathway (Fig. 1) is helpful as a diagnostic tool in them (1). For further evaluation, contrast-enhanced MRI and tumor biopsies should be considered. Also, patients should be informed that even benign tumors (which mainly measure >1 cm) can rupture (20) and cause acute abdomen. In cases of splenomegaly, laparoscopic resection with/without the hand-assisted method should be considered.

Figure Legends

Figure 1

The Heller's Diagnostic and follow-up strategy. (1) *Cyst: imperceptible wall, near-water attenuation (<10

HU), no enhancement. +: Hemangioma: discontinuous, peripheral, centripetal enhancement (findings that are uncommon in splenic hemangiomas). ++: Benign imaging features: homogeneous, low attenuation (<20 HU), no enhancement, smooth mar- gins. §: Evaluate: PET vs. MRI vs. biopsy. ll: Suspicious imaging features: heterogeneous, enhancement, irregular margins, necrosis, splenic parenchymal or vascular invasion, substantial enlargement. ¶: Indeterminate imaging features: heterogeneous, intermediate attenuation (>20 HU), enhancement, smooth margins. #: Follow-up MRI in 6 and 12 months.

Figure 2

Contrast-enhanced computed tomography (CT) scans obtained in case 1

Initial visit: (a) arterial phase; (b) delayed phase

Innumerable splenic tumors were seen.

Six months later: (c) arterial phase; (d) delayed phase

The tumors had enlarged.

Figure 3

Laparoscopic image of the tumors in case 1

Innumerable whitish splenic tumors were seen (arrows).

Figure 4

Contrast-enhanced CT scans obtained in case 2

Initial visit: (a) arterial phase; (b) delayed phase

A solitary splenic tumor (maximum diameter: 2.0 cm) was seen.

Twelve months later: (c) arterial phase; (d) delayed phase

The tumor had enlarged (maximum diameter: 2.5 cm).

Figure 5

Laparoscopic image of the tumor in case 2

A solitary, slightly hard tumor was seen (arrow).

Figure 6

Resected spleen (case 1)

Multiple nodules, consisting of proliferating blood vessels, were found in the spleen.

Figure 7

Resected spleen (case 2)

A cystic nodule was found in the spleen.

Figure 8

Histopathological findings of the resected spleen (case 2)

The tumor consisted of proliferating blood vessels without any epithelial abnormalities. These findings were consistent with hemangioma.

Conflicts of Interest Statement

The authors declare that they have no conflict of interests and that there are no relevant financial disclosures to report.

Authors' contributions

Kazuhiro Hiyama made substantial contributions to the data acquisition and the study conception, design, and interpretation. He was also involved in the initial drafting of the manuscript and critically revising the final manuscript.

The other authors were involved in revising the final manuscript. All of the authors have reviewed the final manuscript and gave their approval to the version to be published. In addition, they agree to be accountable for all aspects of the work, including for ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

List of abbreviations

CA19-9: cancer antigen 19-9, CEA: carcinoembryonic antigen, CT: computed tomography, MRI: magnetic resonance imaging

Consent

Written informed consent was obtained from the patient prior to submission.

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