Solitary Metastasis of Hepatocellular Carcinoma to the Rectus Abdominis 13 Years After the Initial Treatment

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ABSTRACT
Solitary muscle metastasis of hepatocellular carcinoma (HCC) is extremely rare, and late metastasis is also rare. We present a 59-year-old man who had received initial treatment for HCC 13 years previously. Ultrasonography revealed a tumor between the abdominal wall and the liver surface. Tumor resection was performed with suspected intrahepatic metastasis or abdominal wall metastasis of HCC, and the tumor was found to be within the rectus abdominis without an association with the liver. Histologically, the resected material was confirmed to be a muscle metastasis of HCC. We discuss the management of muscle metastasis of HCC.

INTRODUCTION
Hepatocellular carcinoma (HCC) is a common primary hepatic cancer. The most frequent site of extrahepatic metastases is the lungs, followed by the lymph nodes, bones, and adrenal glands.1-3 However, skeletal muscle metastasis is rare, with a reported incidence rate of 0%-1.5%, and solitary muscle metastasis is extremely rare.1,2,4-8 Most recurrences occur relatively early after initial treatment; the reported median duration between initial treatment and extrahepatic metastases is 23.2 months.9 Late metastasis more than 10 years after initial treatment is rare.10,11

CASE REPORT
The patient was a 59-year-old man who had neither hepatitis B virus nor hepatitis C virus infection, with no chronic liver disease or risk factor of HCC. At age 46, he was diagnosed with moderately differentiated HCC and underwent radiofrequency ablation (RFA) twice and transcatheter arterial embolization. After that, he had undergone regular follow-up once in 6 months with abdominal ultrasonography. At age 59, a 24.8 × 14.0-mm tumor was found between the abdominal wall and the surface of liver segment 5 (Figure 1). Subsequent contrast-enhanced computed tomography (CT) imaging revealed that the lesion was contrast-enhanced in the early phase, and the contrast was washed out in the late phase (Figure 2). No other lesions indicating metastases were observed.

Laboratory investigations revealed a normal blood cell count, normal liver enzyme levels, and coagulation. Although the serum α-fetoprotein (AFP) level was normal at 4.5 ng/mL, the protein induced by the vitamin K absence/antagonist-II (PIVKA-II) level was high at 93 mAU/mL. Subsequently, intrahepatic metastasis or abdominal wall metastasis of HCC was established as a differential diagnosis, and tumor resection was planned. During surgery, the tumor was found to be located within the rectus abdominis, not continuous with the liver. The tumor was resected with a margin of approximately 1 cm, surrounding some parts of the rectus abdominis (Figure 3). There was a fistula cranial to the tumor, which seemed to be the needle tract of the former RFA, but it was evidently located away from the resected tumor.

Pathologic examination showed the proliferation of atypical cells with eosinophilic vesicles presenting a pseudo-tubular pattern and immunohistochemically stained with glypican-3, which is compatible with poorly to moderately differentiated HCC. The pathology
is the same as the initial HCC, and muscle metastasis of HCC to the rectus abdominis was confirmed. Three months after the surgery, the serum PIVKA-II level decreased to 20 mAU/mL. Three years after the surgery, there was no evidence of recurrence.

**DISCUSSION**

Skeletal muscle metastasis of HCC is rare, with the reported incidence rate of 0%–1.5%. Although the reason remains unclear, this rarity may be due to several factors, including muscle motion resulting in mechanical tumor destruction, inhospitable muscle pH, and muscles’ ability to remove tumor-produced lactic acid. In our case, the tumor was located away from the scar of the former RFA; hence, we consider it to be hematogenous or lymphogenous metastasis to the rectus abdominis rather than needle track seeding after RFA, whose reported incidence rate is 0%–1.1%.

Solitary muscle metastasis of HCC is extremely rare among all muscle metastases, with only 4 cases being reported. In 2 of those cases, no recurrence was detected in the follow-up period. In 1 case, a pelvic lesion was detected 2 months after the surgery. In another case, intrahepatic recurrence was detected 10 months after the surgery but could be controlled with transcatheter arterial chemoembolization, and recurrence was no longer observed after that. In our case, tumor resection was performed, and no recurrence was detected 3 years after the surgery. Although the management of solitary extrahepatic metastasis remains controversial, surgical resection may be effective for patients with 1 or 2 isolated extrahepatic metastases who exhibit good hepatic functional reserve and general performance status and successful treatment of intrahepatic HCC.

Late recurrence of HCC after initial treatment is rare, and there are only 2 previous reports of extrahepatic recurrence occurring after 10 years. In the first case, the tumor of the right kidney was revealed by CT 12 years after the initial surgery, and surgical resection was performed for suspected renal cell carcinoma. In the second case, the tumor of the right pelvis was revealed by CT and magnetic resonance imaging 28 years after the initial surgery. Hence, surgical resection was performed after the suspicion of degenerative uterine myoma or ovarian fibroma. Late recurrent lesions tend to be diagnosed as a wrong primary tumor because of the rarity of late metastasis of HCC.

In our case, the PIVKA-II level was elevated, whereas the AFP level was normal. Among the 4 cases of solitary muscle metastases previously reported, the serum AFP level did not increase in 2 cases. However, in 1 case, PIVKA-II was elevated. It has been reported that the PIVKA-II level may be a predictive marker for extrahepatic HCC metastases, regardless of AFP level. It is important to measure both AFP and PIVKA-II.
levels to diagnose recurrence and metastasis of HCC. In contrast to CT images, the most common pattern of skeletal muscle metastasis is a focal intramuscular mass with homogeneous contrast enhancement. Among the previously reported cases of muscle metastases of HCC, the common pattern was heterogeneous enhancement in both early and late phases. In our case, the lesion was contrast-enhanced in the early phase, and the contrast was washed out in the late phase, which was compatible with HCC. This finding may be occasionally beneficial in diagnosing HCC metastasis to the skeletal muscle.

We present the case of late solitary muscle metastasis of HCC. It is important to recognize that HCC can metastasize to skeletal muscle even long time after initial treatment. Although it is difficult to distinguish between solitary metastasis of HCC and another kind of tumor, laboratory examination and CT imaging may be useful for diagnosis.

DISCLOSURES

Author contributions: R. Kato wrote the article. A. Sakamoto, T. Noguchi, and S. Matsuda revised the article for intellectual content. H. Terajima approved the final article. A. Sakamoto is the article guarantor.

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REFERENCES


