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Left epigastric isolated tumor feeding with inferior phrenic artery was diagnosed with ectopic hepatocellular carcinoma: a case report

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Abstract

BACKGROUND

Hepatocellular carcinoma (HCC) is one of the most frequent cancers and the main cause of cancer-related death worldwide. Ectopic HCC, an extremely rare sort of HCC, exhibits a wide range of clinical signs and radiographic features, making preoperative identification challenging.

CASE SUMMARY

A 47-year-old man underwent routine abdominal color ultrasonography, which identified an asymptomatic tumor in the left upper abdomen. The patient had no history of hepatitis, did not drink alcohol, and had no family history of cancer. Abdominal contrast-enhanced CT revealed a heterogeneously enhanced lesion between the spleen and stomach that had invaded the diaphragm, with blood supplied by the left inferior phrenic artery. The patient underwent laparoscopic surgery, and HCC was identified by postoperative pathology. Additionally, specific immunohistochemical staining was performed to assess the molecular biological characteristics of the HCC. The patient underwent two rounds of hepatic arterial interventional chemotherapy after surgery. +ADw-span style+AD0AIg-font-size: 14.0pt+ADs- font-family: 'Times New Roman', serif+ADsAIgA+-Abdominal plain and enhanced magnetic resonance imaging

and lung CT 3 mo postoperatively revealed no signs of local recurrence or distant metastasis.

CONCLUSION

Ectopic HCC was generally clinically asymptomatic. Early identification, thorough resection, and systematic surveillance can improve the prognosis.

INTRODUCTION

Hepatocellular carcinoma (HCC) is one of the most prevalent cancers and a major contributor to the global mortality rate from cancer [1]. HCC has a dismal prognosis, with a relative 5-year survival rate of only approximately 18% [2]. Ectopic HCC is an entity of HCC that has no connection to the mother liver and is an extremely rare condition and poses a significant diagnostic challenge before surgery. Ectopic liver tissue, which has an incidence of 0.23% to 0.7%, is hypothesized to be the main source of ectopic HCC [3]. We describe a man with an isolated mass in his left upper abdomen that ultimately proved to be HCC.

CASE PRESENTATION

Chief complaints

A 47-year-old asymptomatic male patient was referred to our hospital due to an mass between the spleen and stomach for more than half a month

History of present illness

The asymptomatic mass was found and suspected as a gastrointestinal stromal tumor during a routine abdominal color ultrasonography.

History of past illness

He had no medical history, no history of viral hepatitis, and no history of smoking or consuming alcohol.

Personal and family history

There is no history of malignancy in the family.

Physical examination

Physical examination revealed no obvious mass or tenderness. Bowel sounds were within an acceptable range, while rectal palpation and ascites examinations were negative.

Laboratory examinations

On laboratory examination, the only specific abnormality was an elevated α-fetoprotein (AFP) concentration (194.38 ng/mL). The results of other laboratory tests, namely hemoglobin, gamma glutamyl transferase, alkaline phosphatase, anti-hepatitis C virus, HBV surface antigen, carcinoembryonic antigen, and cancer antigen 19–9, were within normal limits.

Imaging examinations

Abdominal ultrasonography revealed a heterogeneous mass measuring approximately 6 × 9 cm between the spleen and stomach. Contrast-enhanced CT showed that the shape and size of the liver were normal, the edges were smooth, the proportion of each lobe was balanced, and no obvious abnormal density shadows were identifiable in the liver parenchyma. A soft tissue mass was observed in the splenic and gastric spaces, with a CT value of 48 HU and size of 6.3 × 9.3 cm. Enhanced CT showed uneven enhancement (Figure 1). CT values during the arterial, portal, and extended stages were 71-125 HU, 82-112 HU, and 72-85 HU, respectively (Supplementary Figure 1). The tumor feeding artery originated from the left inferior phrenic artery.

FINAL DIAGNOSIS

An ectopic HCC with diaphragmatic involvement was eventually diagnosed.

TREATMENT

The tumor was located in the left subdiaphragm, which was a challenge to access with open surgical operations. Therefore, we performed a laparoscopic surgical operation. During the operation, no ascites was found, and the liver was normal in size, color, and consistency. A tumor with an intact envelope was found between the greater curvature of the stomach and the spleen. It was not related to the hepatic parenchyma and had invaded the diaphragm (Figure 2). We removed part of the diaphragm to ensure that the tumor was entirely resected because the tumor had direct diaphragmatic involvement. Preventive hepatic arterial infusion chemotherapy was administered to the patient twice, 1 mo apart. The potential for intrahepatic microneoplasms, such as micrometastases or a microprimary tumor with metastasis to the left upper abdomen, was ruled out by hepatic arteriography (Figure 3).

OUTCOME AND FOLLOW-UP

Regular monitoring and follow-up were performed after surgery as advised during a multidisciplinary team meeting that examined AFP (Figure 4), lung and skull CT scan, and liver magnetic resonance imaging (Figure 5).

Hematoxylin and eosin staining, and immunohistochemistry were used to thoroughly assess the tumor's type and immunological markers (Figure 6). Capsule formation is related to better tumor cell differentiation and tumor biological characteristics compared with no capsule formation, and the pathology results of this patient suggested a well-to-moderately differentiated adenocarcinoma. Immunohistochemical study revealed the following: hepatocyte antigen (+), arginase-1 (+), AFP (+), inhibin-α (−), cytokeratin (CK) (+/−), GPC-3 (+), Ki-67 (hot regions: 5%), CK19 (−), CK8/18 (−), and hepatocyte paraffin-1 (+). To determine whether the patient could benefit from immunotherapy, we also performed programmed death-ligand 1 testing. However, the results were negative. Consequently, intensive postoperative immunotherapy was not administered.

DISCUSSION

HCC is a common malignancy of the digestive system with high morbidity and mortality worldwide. However, only 27 cases of ectopic HCC have been reported as of 2022[4]. The risk factors for ectopic HCC are unrelated to those for typical HCC (such as chronic viral hepatitis B and C; alcohol abuse or alcoholic steatohepatitis; and nonalcoholic fatty liver disease or nonalcoholic steatohepatitis). Ectopic HCC may be caused by problems with biliary drainage, insufficient arterial blood flow, and insufficient venous drainage [3,5]. The prolonged exposure to these carcinogenic factors promotes carcinogenesis. In the present case, the patient had no substantial coexisting conditions or HCC risk factors, and the mother liver was not cirrhotic. The most common extra-hepatic feeding arteries of HCC were the inferior phrenic artery (IPA) [6]. Our patient presented with an isolated mass in the left upper abdomen, invaded the diaphragm and fed by the left IPA.

Owing to the distinct features of ectopic HCC, patients present with a wide spectrum of clinical signs and radiological presentations, which makes it challenging to obtain a preoperative diagnosis [7]. The case that we described was an isolated tumor with no connection to the liver and supplied by diaphragmatic arteries, which made preoperative diagnosis challenging. Anatomically, part of ectopic HCC shows a significantly better overall survival than that for HCC in the orthotopic liver because of encapsulated and the possibility of wide resection margins with less vascular invasion [8]. Our patient had well-to-moderately differentiated histological characteristics, with a low Ki-67 Labeling index, which may indicate a highly favorable overall survival compared with other histological subtypes.

Some HCCs can attach to and potentially infiltrate nearby tissues, such as the diaphragm, which presents therapeutic difficulty. During the preoperative examination, whether the diaphragm is actually invaded by HCC is difficult to identify; therefore, a postoperative pathological diagnosis or intraoperative frozen pathology is necessary to evaluate whether the diaphragm has been invaded or is just showing diaphragmatic

fibrous adhesion [9]. Because there is a risk of tumor rupture, tumor spread, and bleeding when the diaphragm is separated from a tumor that has gross diaphragmatic involvement, the grossly involved section of the diaphragm must be excised to ensure radical cure [10]. However, whether diaphragmatic excision is necessary and beneficial for individuals with subclinical adhesion and a smooth, intact tumor capsule is debatable. In the present case, part of the diaphragm was resected to ensure radical resection without postoperative complications. However, extrapolating general implications from a single instance might be challenging. Laparoscopic techniques are widely used and well recognized by the majority of liver surgeons owing to reduced patient stress and rapid postoperative recovery [11]. However, the use of these techniques in HCC with diaphragmatic involvement has not been widely reported [12]. We have found that this approach is reliable and efficient, helps reduce surgical complexity and operation time, and enhances surgical safety; however, high-quality clinical trials are required. With advanced-stage HCC, comprehensive postoperative care is required, including preventive transcatheter arterial chemoembolization, anti-HBV therapy, immunotherapy, and targeted therapy. Close, frequent monitoring and follow-up are also necessary following surgery, as with resection or radiofrequency ablation of recurring foci [13]. In the present case, we performed two sessions of preventive transhepatic arterial perfusion chemotherapy. To date, there has been no evidence of recurrence.

CONCLUSION

We reported a patient with an isolated tumor between the spleen and the stomach that was diagnosed as ectopic HCC. Excellent curative results were obtained with treatment.

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