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Surgical resection of splenic metastasis from the adenosquamous gallbladder carcinoma: A case report

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ABSTRACT

INTRODUCTION: Splenic metastasis of gallbladder carcinoma is extremely rare. Specific anatomical, histological, and functional properties of spleen are believed to be responsible for the rarity of solitary splenic metastasis.

PRESENTATION OF CASE: We present the case of a 62-year-old female who developed metachronous splenic metastasis of adenosquamous carcinoma of the gallbladder. We performed central bisegmentectomy of the liver for gallbladder carcinoma. The patient subsequently presented 3 months later with isolated splenic metastasis and liver metastasis. Splenectomy and partial hepatectomy was performed at this time. Histological examination confirmed metastatic adenosquamous carcinoma of the gallbladder. No signs of recurrence were observed at 3 months after the second surgery.

DISCUSSION: Although splenectomy provides a potential means of radical treatment in patients with isolated splenic metastases, it should be performed with caution as splenic metastatic lesions may represent the initial clinical manifestation of systemic metastases at multiple sites. In this case, radical surgery was performed following the confirmation of no new unresectable metastatic lesions or systemic dissemination.

CONCLUSION: This is the first report on the adenosquamous splenic metastasis from the gallbladder carcinoma. Curative resection may be the treatment of choice for prolonging survival in patients with the splenic metastasis of gallbladder carcinoma.

1. Introduction

Splenic metastases are rare, comprising 0.96% of reported cases of metastatic cancer [1]. Specific anatomical, histological, and functional properties of the spleen are believed to be responsible for the rarity of solitary splenic metastasis. As majority of splenic metastases are detected concurrently with multi-organ metastases, isolated splenic metastasis is extremely rare. Gallbladder (GB) carcinoma commonly metastasizes to the liver and regional lymph nodes and rarely the spleen. Adenosquamous carcinoma of GB is a relatively rare (<10%) type of gallbladder carcinoma, defined as being comprised of 25–99% squamous cells [2,3].

In this study, we report a case of splenic metastasis of adenosquamous carcinoma of GB occurring 3 months after initial curative treatment which was treated by splenectomy.

2. Presentation of case

We report the case of a 62-year-old female who presented with right hypochondralgia and was subsequently diagnosed with gallbladder cancer by abdominal ultrasonography before referral to our hospital for possible surgery. Computed tomography (CT) revealed an irregular gallbladder mass involving the liver (Fig. 1). A preoperative diagnosis of stage T3N0M0 gallbladder carcinoma was made according to the UICC classification [4]. Serum levels of tumor biomarkers, including carcinoembryonic antigen and carbohydrate antigen 19-9 (CA19-9), were within the normal limits. However, squamous cell carcinoma antigen (SCC) was higher than the normal range (6.5 ng/ml; reference values, 0–1.5 ng/ml). Central bisegmentectomy of the liver with partial resection of transverse colon, as performed as laparotomy, revealed liver metastasis in segment 8 and invasion into the transverse colon. Lymph node dissection at

Abbreviations: GB, gallbladder; CT, computed tomography; MRI, magnetic resonance imaging; FDG-PET, fluorodeoxyglucose positron emission tomography; SSC, squamous cell carcinoma antigen.

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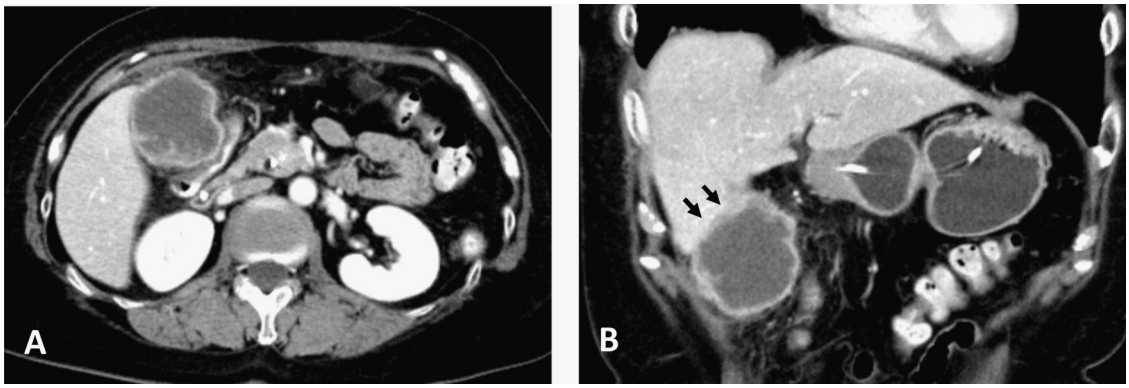

Fig. 1. Abdominal computed tomography reveal the gallbladder carcinoma.
(A) The axial view of contrast-enhanced CT. (B) The coronal view of contrast-enhanced CT. Contrast-enhanced CT demonstrating increased gallbladder wall thickness and a low density irregular mass within the gallbladder. The mass was diagnosed as gallbladder cancer with invasion into the liver (arrows).

the hepatoduodenal ligament and along the common hepatic artery was also performed.

Macroscopically, the GB tumor was 8.0 cm in size with massive invasion into the liver and transverse colon. Histopathological examination of the specimen demonstrated adenosquamous carcinoma with liver metastases in segment 8 and lymph node metastases along the common bile duct. Therefore, the final pathological stage was determined as pT4N1M1, pStage IV, according to the UICC system [4]. Serum SCC levels decreased to 0.6 ng/ml during the early postoperative period. Two courses of adjuvant chemotherapy consisting of TS-1 80 mg/day for 14 days were administered with a 7-day interval.

Three months after initial surgery, serum SCC levels elevated to 8.7 ng/ml. Enhanced CT imaging revealed a low density mass in the spleen (Fig. 2A). Fluorodeoxyglucose positron emission tomography (FDG-PET) demonstrated abnormal FDG accumulation in the spleen (Fig. 2B). Other remote organ metastases were not observed on either CT or FDG-PET imaging. The previous history of GB carcinoma contributed to a presumptive diagnosis of metachronous isolated splenic metastases.

Two courses of intravenous chemotherapy consisting of gemcitabine (1200 mg) and cisplatin (30 mg)/day were administrated due to early metastasis after the initial surgery. However, serum SCC levels gradually increased to 65.8 ng/ml. Five months after the initial surgery, CT imaging revealed an enlarging splenic tumor and new liver metastasis in segment 6, indicating a poor response to chemotherapy (Fig. 2C). No other intra-abdominal organ metastases or peritoneal dissemination were observed at this time. Therefore, splenectomy and partial liver resection were performed for the treatment of splenic and liver metastases. Upon laparotomy, intraoperative lavage cytology was negative and no recurrence of nodular masses was observed except in the spleen and liver. As the splenic tumor had invaded into the left diaphragm, combined partial resection of the left diaphragm was performed.

Macroscopically, the splenic tumor was solitary and homogeneuous. The cut surface of the resected specimen revealed a whitish splenic mass with a diameter of 6.7 cm (Fig. 3). Histopathologically, the splenic and liver tumors were found to be adenosquamous carcinoma, consistent with metastasis of the primary GB carcinoma (Fig. 4). The postoperative course was uneventful. SCC levels decreased to within normal limits. The patient was discharged on postoperative day 13. Chemotherapy consisting of TS-1 or gemcitabine and cisplatin was not effective. Therefore, we administered adjuvant chemotherapy consisting of docetaxel and nedaplatin, which we expected to be effective. There was no sign of recurrence at 3 months after the second surgery.

3. Discussion

Splenic metastases from non-hematologic malignancies account for only 0.96% of the metastatic carcinoma and 2.9–4.4% of the autopsied carcinoma specimens [5,6]. In majority of cases, the spleen is involved as a part of diffuse carcinomatosis with the presence of splenic metastases usually indicating widespread tumor dissemination [7,8]. Very few splenic metastases are observed as isolated splenic lesions, synchronous or metachronous to the primary tumor. There have been few case reports and reviews of isolated splenic metastases from carcinoma of the colorectum, stomach, kidney, or ovary [9–14]. Splenic metastasis from GB cancer is extremely rare. Taki et al. first reported isolated splenic metastasis from adenocarcinoma of GB [15]. Therefore, this presents the second reported case of the splenic metastasis of GB successfully treated by surgical resection. According to histopathological diagnosis, to the best of our knowledge, this is the first report of splenic metastasis of adenosquamous cancer of the gallbladder.

The rarity of splenic metastases is believed to be due mechanical factors and the splenic microenvironment [16]. Mechanical factors related to the spleen, such as constant blood flow through the spleen, rhythmic contraction of the splenic capsule, the sharp angle of the splenic artery after branching from the celiac trunk, and the lack of afferent lymphatic vessels, may protect against tumor cells infiltration and the development of metastases [17]. Further, the splenic microenvironment may inhibit cancer cell proliferation [17,18]. Although the precise mechanisms underlying the inhibition of cancer cell growth has yet to be fully elucidated, the immunological environment of the spleen may be particularly hostile to tumor cells.

Adenosquamous carcinoma of GB is a relatively rare type of gallbladder carcinoma associated with worse prognosis. Primary GB carcinoma includes the following histological types in a decreasing order of incidence: adenocarcinoma, adenosquamous carcinoma, squamous cell carcinoma, and oat cell carcinoma [19]. In GB carcinoma, tumor stages of adenosquamous and squamous cell carcinoma are significantly advanced at presentation, compared to adenocarcinoma [3,20,21]. The overall prognosis of adenosquamous GB carcinoma appears to be worse, with most patients dying within few months of diagnosis [2]. However, curative surgical resection of adenosquamous or squamous cell carcinoma might result in disease-free survival rates comparable to those for adenocarcinoma [3,20,21].

A metastatic origin should be suspected for all isolated splenic tumors found during oncologic follow-up. Serum tumor marker levels reportedly have predictive value in detecting isolated splenic