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# Multimodal Treatment of Hepatic Metastasis in the Form of a Bile Duct Tumor Thrombus from Pancreatic Acinar Cell Carcinoma: Case Report of Successful Resection after Chemoradiation Therapy

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## Key Words

Acinar cell carcinoma · Hepatic metastasis · Bile duct tumor thrombus

## Abstract

Pancreatic acinar cell carcinoma (ACC) is a rare tumor, and its pathophysiology has not been well understood. Treatment strategies for hepatic metastasis originating from ACC remain controversial. We report the case of a 66-year-old woman who had undergone total pancreatectomy from ACC 7 years prior to clinical presentation. Contrast-enhanced computed tomography imaging revealed a tumorous lesion measuring 7 cm in length and 1 cm in diameter and extending along the intrahepatic bile duct (B6), which showed mild enhancement in the early phase and modest washout in the late phase. This lesion was diagnosed as hepatic metastasis primarily in the form of a bile duct tumor thrombus originating from the prior ACC by the pathological evaluation of the fine needle biopsy specimen. The patient underwent preoperative gemcitabine-based chemoradiation therapy followed by subsequent surgical resection, which included subsegmentectomy (S6) of the liver and complete removal of the bile duct tumor thrombus. The patient has had no recurrence during the past 8 months since her last surgery. Multimodal treatment including preoperative chemoradiation therapy might be beneficial especially for marginally resectable cases of ACC.

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## Introduction

Acinar cell carcinoma (ACC) of the pancreas is a rare tumor that is estimated to account for 1–2% of all pancreatic exocrine tumors [1, 2]. Because of the lack of clinical experience with ACC, its pathophysiology is not well understood. According to previous reports, ACC generally shows expansive growth without invasion into the plexus, portal vein, bile duct and pancreatic duct [3, 4]. To our knowledge, no previous reports have described hepatic metastasis from ACC primarily in the form of a tumor thrombus. We encountered a very rare case of hepatic metastasis with an unusual clinical presentation: a bile duct tumor thrombus (BDTT) from pancreatic ACC. This tumor was successfully treated using a multimodal approach that included resection.

## Case Report

A 66-year-old woman had undergone total pancreatectomy due to pancreatic ACC (T3N0M0, stage IIa) 7 years prior to clinical presentation [5]. Computed tomography (CT) imaging showed a tumorous lesion measuring 7 cm in length and 1 cm in diameter and extending along the intrahepatic bile duct (B6). During CT imaging, the lesion showed mild enhancement in the early phase and modest washout in the late phase (fig. 1a). CT arteriography (CTA) showed early enhancement, which corresponded to the tumorous lesion detected by CT (fig. 1b). CT arterial portography (CTAP) showed a partial perfusion defect (fig. 1c). No other lesions were detected by either CTA or CTAP. <sup>18</sup>F-fluorodeoxyglucose positron emission tomography/CT (FDG-PET/CT) showed high FDG uptake within the corresponding area (fig. 1d). The tumor was primarily confined to the intrahepatic bile duct (B6) without definitive mass formation in the hepatic parenchyma. Tumor markers, including CEA, CA19-9 and DUPAN-II, were all within normal limits. Since pathological evaluation of the fine needle biopsy specimen from the BDTT revealed histological features that were compatible with those of the patient's ACC resected 7 years before, the lesion was diagnosed as a hepatic metastasis primarily in the form of a BDTT originating from the prior ACC.

We initiated chemoradiation therapy (CRT), consisting of a total radiation dose of 50 Gy and the intravenous administration of gemcitabine (1,000 mg/m<sup>2</sup>) concurrently on days 1, 8, and 15 during each 4-week cycle. Administration of gemcitabine was continued after completion of the radiation therapy. Although a modest effect was observed 13 weeks after the initiation of CRT, with a decrease in blood flow and size of the BDTT (fig. 2a), the lesion showed an increase in size 19 weeks after the initiation of CRT (fig. 2b). Thus, surgical resection, including subsegmentectomy (S6) of the liver and complete removal of the BDTT, was performed. Pathological evaluation of the resected specimen confirmed a hepatic metastasis primarily consisting of a BDTT that originated from an ACC of the pancreas (intraductal polypoid growth variant). The detailed pathological evaluation of the BDTT and the comparison of this tumor's characteristics with those of the primary pancreatic lesion that was resected 7 years before are presented in a separate report [6]. The patient is currently alive without any recurrence 8 months after the second operation.

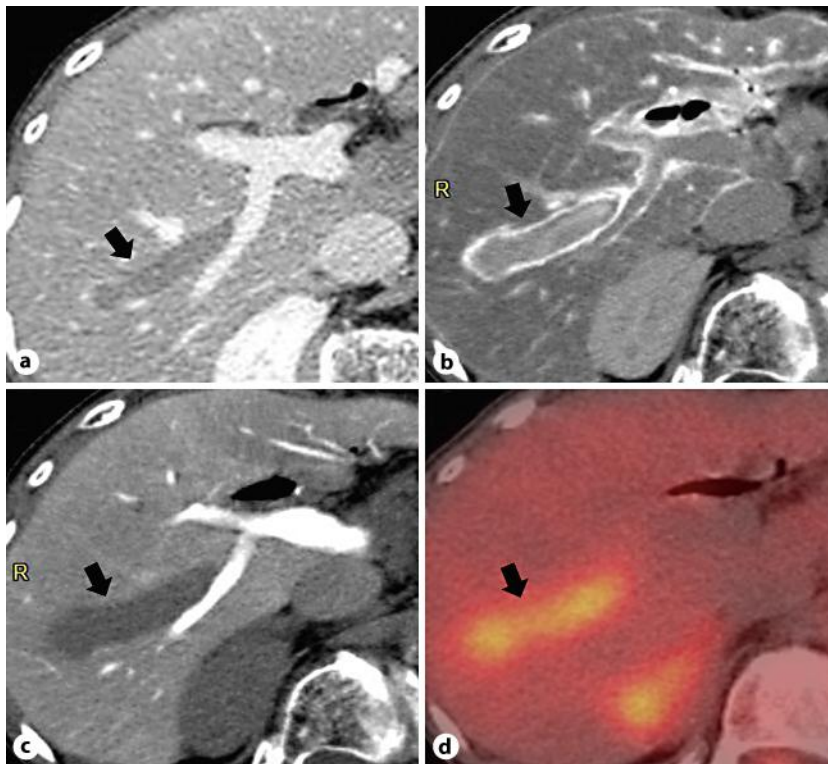
## Discussion

Although our experience is only anecdotal, the case reported here provides several novel observations in regard to the clinical features associated with pancreatic ACC. First, it underscores the need for a long-term follow-up for pancreatic ACC cases, even after curative resection. This case involved a liver metastasis that was detected more than 5 years postoperatively, which is unique as hepatic metastases normally occur within a relatively short period following resection of the primary lesion [7, 8]. Second, aggressive surgical resection combined with CRT for hepatic metastasis from pancreatic ACC should be considered as a treatment option in selected cases. Surgical

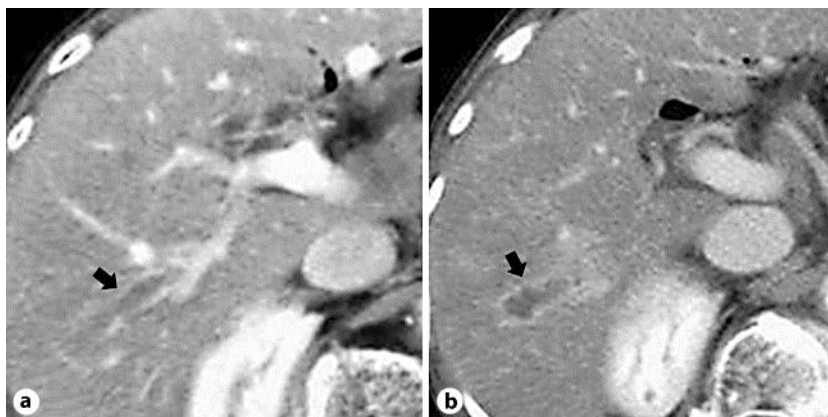
resection is recognized as a standard treatment strategy for localized ACCs [9]. However, the treatment strategy for metastatic or recurrent ACC remains controversial even though various treatment options, including chemotherapy, CRT and surgical resection, have been reported [7–8, 10–12]. Suzuki et al. [8] reported a case of hepatic metastasis from ACC that was successfully treated with an aggressive surgical approach. The advantage of CRT prior to surgical resection for hepatic metastasis from ACC is unclear. However, a preoperative CRT strategy might be beneficial especially in marginally resectable cases because a partial response to CRT was observed in the case presented here. Third, although multimodal approaches may be useful for selected ACC cases [10–12], careful repeated re-evaluations during the preoperative treatment period are necessary to improve the chances for a curative resection as demonstrated by our current case, in whom the BDTT showed regrowth after a partial response to CRT. Further studies with a larger number of patients are required to fully understand the pathophysiology of ACC and to evaluate the benefits of multimodal treatment strategies for recurrent ACC.

#### Disclosure Statement

The authors have no potential conflict of interest.



**Fig. 1.** CT and FDG-PET/CT. **a** Enhanced CT showed an area of low density along the intrahepatic bile duct (B6) (arrow). **b** CTA showed early enhancement (arrow). **c** CTAP revealed a partial perfusion defect which corresponded to the area of low density detected by CT (arrow). **d** FDG-PET/CT showed high FDG uptake within the corresponding area (arrow).



**Fig. 2.** Changes to the metastatic lesion after CRT. **a** The tumor shrunk slightly 13 weeks after the initiation of CRT (arrow), but a CT performed 6 weeks after the initial evaluation revealed that the tumor had re-grown (**b**, arrow).

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