



Metastatic Cholangiocarcinoma Presenting as Colonic Obstruction

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ABSTRACT

Cholangiocarcinoma (CCA) is a biliary epithelial cancer conferring a grim prognosis. Metastatic recurrence of CCA is often discovered because of symptoms stemming from the site of metastasis. It is rarely reported that colonic obstruction serves as the presenting symptom of disease recurrence. We report the unusual discovery of malignant colonic obstruction because of metastatic CCA. Our report highlights the benefit of minimally invasive colonic stenting for palliation yielding immediate relief of pain while avoiding untoward sequelae from high-risk surgical intervention.

KEYWORDS: cholangiocarcinoma; metastatic cholangiocarcinoma; colonic obstruction; colonic stenting

INTRODUCTION

Cholangiocarcinoma (CCA) is a biliary epithelial cancer with a grim prognosis. Diagnosis usually stems from evaluation of downstream effects such as jaundice or other signs of biliary obstruction.¹ Seldom has colonic obstruction been documented to be the presenting symptom prompting the discovery of metastatic disease. We report such a case, in which a patient with a history of CCA was found to have metastatic disease manifesting as a sigmoid colonic stricture.

CASE REPORT

A 67-year-old white woman presented with complaints of abdominal bloating, distention, and constipation for 2 weeks. Her medical history was significant for perihilar cholangiocarcinoma diagnosed 4 years earlier, which was discovered after evaluating for the etiology of asymptomatic jaundice. Her last colonoscopy was 8 years earlier and featured no abnormal colonic findings. She was status-post several interventions including left hepaticojejunostomy/Roux-en-Y and a biliary drain placement for the perihilar cholangiocarcinoma, with positron emission tomography postprocedurally demonstrating no colonic involvement. She was treated with adjuvant chemoradiation that was discontinued after a duodenal channel ulceration was discovered. Recovery was complicated by a hepaticojejunostomy fistulation to her abdominal wall. Two years after the initial diagnosis, she experienced an umbilical cutaneous metastasis, which was surgically resected. Computed tomography (CT) of the abdomen at that time confirmed no additional involvement.

On this visit to the emergency department, vital signs were notable for a heart rate at 107 bpm, and physical examination revealed a distended abdomen that was significantly tender to palpation but lacked peritonitic findings. Serologic testing was notable for an elevation in total bilirubin to 1.7 mg/dL. CT imaging revealed pelvic ascites and retained stool material, despite oral laxatives and enemas. Repeat gastrografin enemas were unsuccessful, and a diagnostic flexible sigmoidoscopy was pursued. During this procedure, a smooth-appearing circumferential sigmoid stricture with edematous mucosa was visualized. The stricture measured 1 cm (length) × 3 mm (inner diameter) and was visualized 15 cm from the anal verge. This area was traversed with a neonatal scope, and biopsies revealed tissue that was morphologically similar to the original liver resection (CK7+, CK20–), confirming metastatic cholangiocarcinoma. Palliative colonic stenting was recommended after discussion with surgical oncology because of the high propensity for surgical complications, given the patient's ascites (Figures 1 and 2).

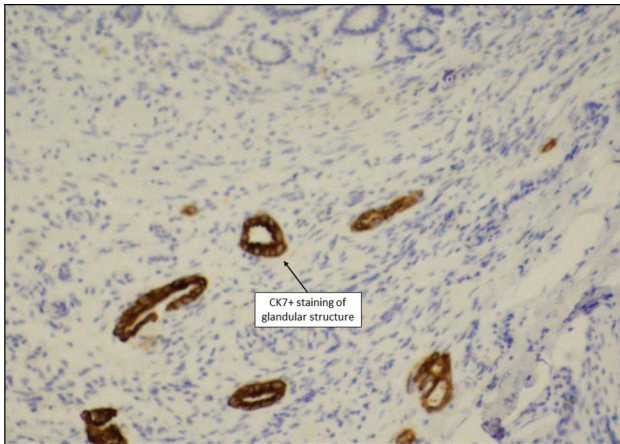


Figure 1. Histopathologic stain of stricture biopsy confirming CK7 positivity of glandular structures in the colonic muscularis mucosa; recurrent cholangiocarcinoma presentation (200 \times).

The therapeutic procedure ensued, with a Boston-scientific uncovered 22 \times 90-mm colonic stent consequently being placed across the stricture. Decompressive liquid stool was seen immediately after stent deployment, indicating successful placement. A diet was resumed on postoperative day 1 (Figure 3–5).

The remaining hospital course was complicated by a biliary drain blockage requiring exchange and upsizing, and a percutaneous cholecystostomy tube tract vascular pseudoaneurysm leading to

recurrent hematochezia and hemobilia, which required embolization of the right hepatic artery at the hepatic hilum. She underwent several therapeutic paracenteses because of repeat ascites accumulation, and CT imaging revealed a portal vein occlusion. A tunneled catheter was placed for palliative management of the ascites, and she was discharged under hospice care (Figure 6).

DISCUSSION

Cholangiocarcinoma is a rare malignancy of the biliary epithelium. Metastatic dissemination of perihilar disease most often occurs through lymphatic spread, whereas intrahepatic CCA spreads through the hepatic venous system.¹ Metastatic spread to the colon is a seldom documented phenomenon, of which we located 10 published case reports.^{2–12} Of these, only 2 feature malignant colonic obstruction.^{10,13}

Literature describing CCA metastasis features both synchronous and dyssynchronous presentations. In many of the synchronous presentations, obstructive biliary symptoms prompt further workup. Wakahara describes the concurrent presentation of a hepatic hilar mass and a colonic mucosal mass that were discovered because of biliary obstruction.¹² This report mentioned hepatic perineural invasion and reported the colonic mucosa was intact, both findings consistent with our case presentation. Vabi et al featured the discovery of colonic metastasis after biliary strictures were investigated through

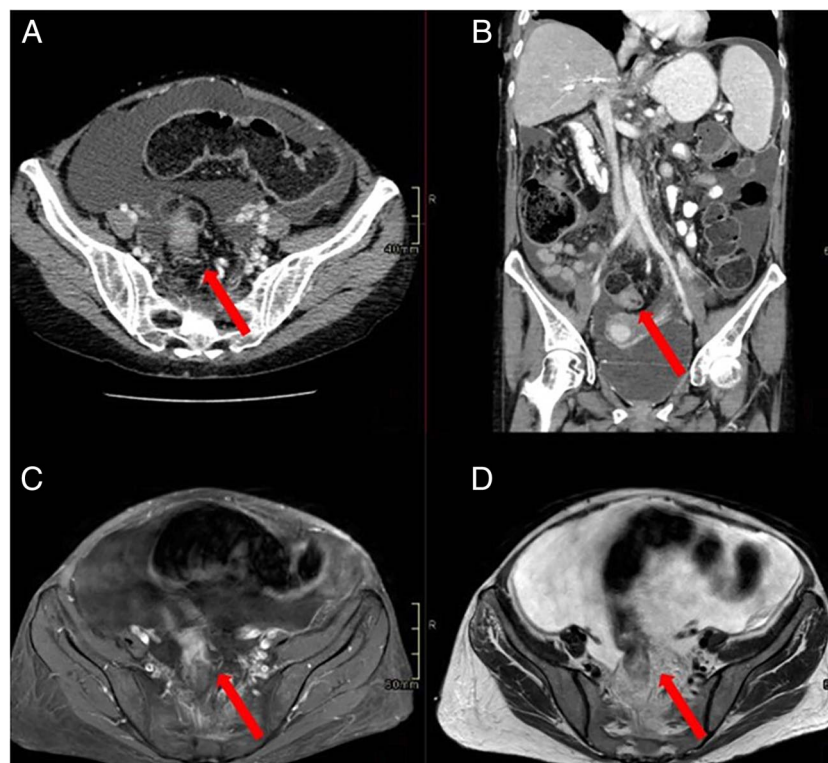


Figure 2. CT image of sigmoid stricture in transverse plane (A) and coronal plane (B); magnetic resonance imaging (musculoskeletal protocol) of sigmoid stricture in T-1 fat-saturation postcontrast imaging (C) and T-2 weighted imaging (D). CT, computed tomography. Arrows point to area of stricture.

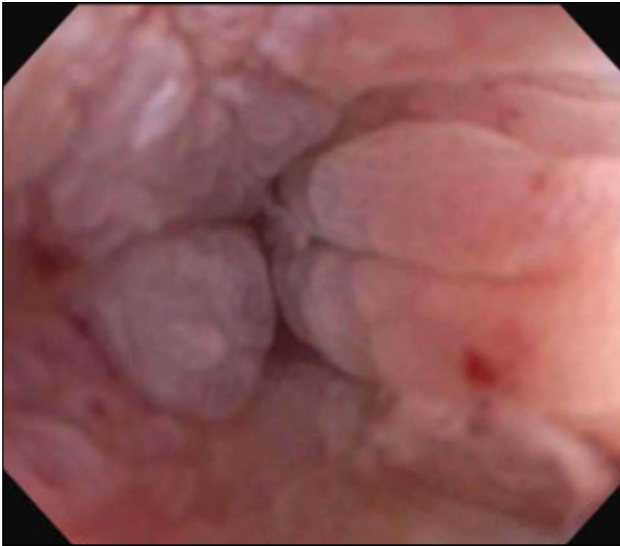


Figure 3. Endoscopic view of colonic stricture.

colonoscopy, which discovered diffuse colonic thickening was observed without a distinct mass lesion.¹¹ Dyssynchronous presentations include cases of metastatic recurrence to the colon 5–6 years after primary cancer resection.^{2,9,10}

Although the symptomatic profile of CCA metastasis varies by location of metastasis, hematochezia has been infrequently mentioned. In one case, hematochezia was ultimately attributed to ulcerative friability of colonic metastases.⁸ Our case featured hematochezia attributed to a vascular pseudoaneurysm, but bleeding because of her mucosal friability cannot be entirely excluded.

The variable expression of cytokeratin (CK) immunohistochemical staining has been used to differentiate colonic adenocarcinoma

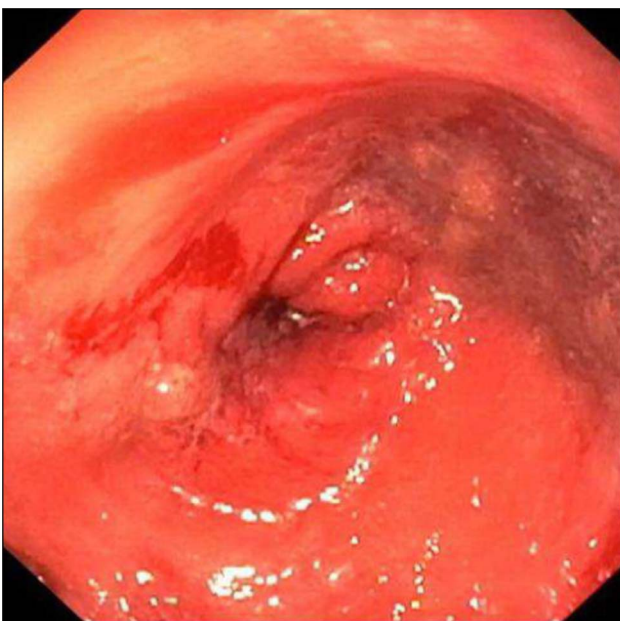


Figure 4. Endoscopic view of colonic stricture.

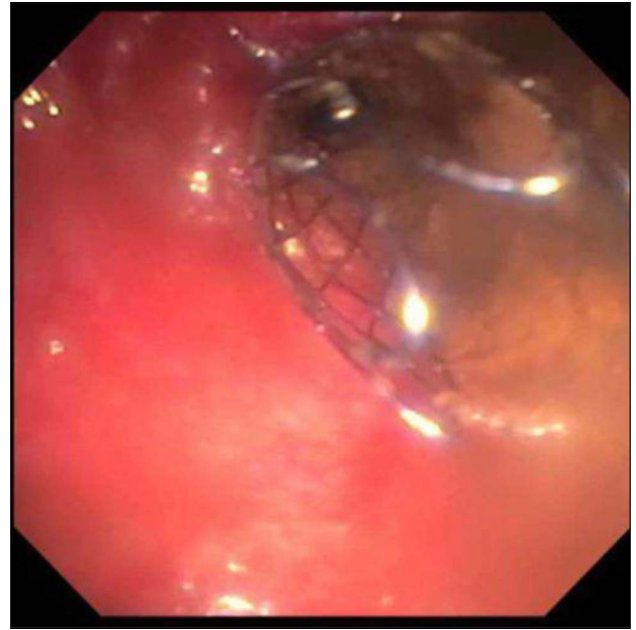


Figure 5. Endoscopic view of liquid stool passing through an uncovered, 22 × 90-mm WallFlex colonic stent.

and CCA. Primary colorectal carcinomas almost unanimously feature CK20 positivity, whereas a pattern of CK7+/CK20– is highly indicative of peripheral CCA.¹⁴ Our case corroborates this, featuring strong CK7 positivity and CK20 negativity. There has also been discussion correlating elevations in CA 19-9 serum level to CCA recurrence. Tokodai et al correlated the discovery of a hepatic flexure stricture to CCA 6 years after surgical removal of the primary intrahepatic lesion after a CA 19-9 elevation was seen.¹⁰ Our patient’s levels of CA 19-9 were variable, and no elevation was correlated with the time of presentation. Readings within the 6 months preceding admission,



Figure 6. Fluoroscopic image of deployed stent traversing colonic stricture. Arrows outline deployed stent.

in U/mL, were 180 (1 month before), 143 (3 months before), and 270 (6 months before).

Treatment of CCA historically consists of surgical intervention. Combining surgery with chemotherapy and/or immunotherapy has been theorized to improve survival.¹⁵ Unresectable tumors usually require decompressive biliary stenting or drainage for symptomatic management and to facilitate hepatically metabolized chemotherapy for disease burden management.^{1,16} Favorable prognosticators include intrahepatic primary tumors, clear surgical margins, and well-differentiated histology.¹⁷ Our patient's initial tumor histopathology featured poor differentiation and perineural involvement at time of primary resection, conferring poorer prognosis. In patients with perihilar involvement who underwent surgical resection with clean margins, median survival has been documented at 30 months, and the 5-year survival rate is estimated at 30%.¹⁷ Although our patient did initially undergo chemotherapy and surgical excision, chemotherapy was discontinued because of adverse effects 4 years before recurrence. Palliative colonic stenting optimized surgery-free longevity to 9 months postprocedurally, and she survived 61 months from her original diagnosis.

In conclusion, our case highlights the exceedingly rare clinical phenomenon of colonic obstruction because of cholangiocarcinoma recurring several years after primary tumor excision.

DISCLOSURES

Author contributions: Y. Protopapadakis wrote and revised the article. K. Lamm and J. Baber reviewed and edited the article. J. Baber is the article guarantor.

Financial disclosure: None to report.

Previous presentation: ACG Annual Scientific Meeting 2022; October 24, 2022; Charlotte, North Carolina.

Informed consent was obtained for this case report.

Received July 12, 2023; Accepted November 16, 2023

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