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CASE REPORT | COLON

Postpolypectomy Bleeding Gone Wrong: Primary Colonic Epithelioid Angiosarcoma

Rishi Bolla, MD[1], Carolina Martinez, MD[2], David Metter, MD[3], and Jonathan Hilal, MD[1]

[1]*Department of Internal Medicine, Division of Digestive Diseases and Nutrition, University of South Florida, Morsani College of Medicine, Tampa, FL*
[2]*Department of Surgery, University of South Florida, Morsani College of Medicine, Tampa, FL*
[3]*Department of Pathology and Cell Biology, University of South Florida, Morsani College of Medicine, Tampa, FL*

ABSTRACT

Epithelioid angiosarcoma is an aggressive form of angiosarcoma, and primary colonic tumors are extremely rare. We present the case of a 60-year-old man who presented with what initially appeared to be postpolypectomy bleeding. After undergoing repeat endoscopy, he was found to have epithelioid angiosarcoma of the transverse colon, and imaging further confirmed that it was the primary tumor site. Our patient underwent segmental resection of his transverse colon. The patient's initial presentation as postpolypectomy bleeding and his continued cancer-free survival after a relatively limited surgery are unique features not previously seen.

KEYWORDS: colon cancer; angiosarcoma; polypectomy; bleeding

INTRODUCTION

Epithelioid angiosarcoma is an aggressive tumor consisting of malignant endothelial cells bearing an epithelioid appearance.[1] It can arise from anywhere in the body, but primary colonic epithelioid angiosarcoma (PCEA) is exceptionally rare, with only a few cases reported in the literature. We present a case of PCEA in which the patient underwent resection of his tumor and, at the time of writing, is alive and free of cancer.

CASE REPORT

A 60-year-old man presented to our hospital with complaints of hematochezia and symptomatic iron deficiency anemia. A year prior, after a positive fecal immunochemical test, he underwent a diagnostic colonoscopy in which a reported 20 mm polyp in the distal transverse colon was found and removed en bloc with hot snare cautery. Pathology revealed it to be benign colonic mucosa. Over the course of several months postprocedure, he began to have intermittent, persistent rectal bleeding. One month before presentation, he underwent a repeat colonoscopy which revealed active bleeding coming from the prior polypectomy site in the transverse colon. This was treated with bipolar diathermy with successful hemostasis, and the wall opposite to the area was tattooed for future localization. Just a few days after this colonoscopy, he began to experience near daily hematochezia with almost every bowel movement which prompted him to present to our institution for a second opinion.

Initial workup revealed that he had significant iron deficiency anemia (hemoglobin 7.2 g/dL, ferritin 7.6 ng/mL) and repeat colonoscopy revealed a submucosal, ulcerated, nonobstructing mass opposite the tattoo site in the distal transverse colon (Figure 1). The mass was noncircumferential and approximately 2 cm in maximum diameter. Bite-on-bite biopsies were taken from the ulcerated portion of the lesion for histology, and 2 hemostatic clips were placed, achieving successful hemostasis.

Pathology from the endoscopic biopsies revealed that the mass showed architecturally atypical vascular proliferation with vascular spaces lined by plump CD31 and D2-40 positive epithelioid endothelial cells, consistent with epithelioid angiosarcoma (Figure 2).

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Correspondence: Rishi Bolla, MD (bollar@usf.edu).

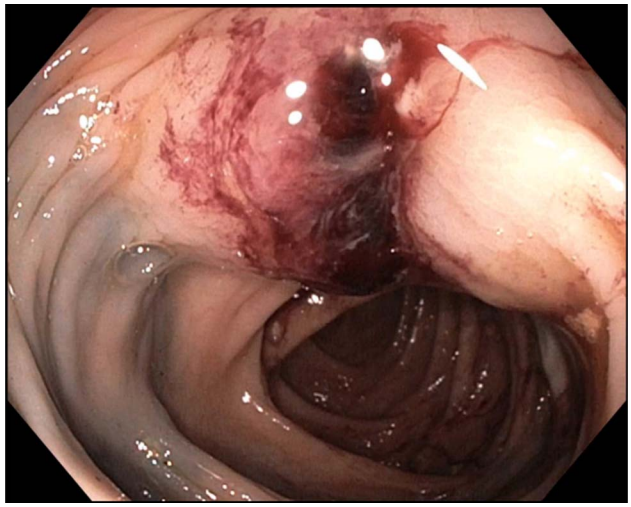

Figure 1. Endoscopically visualized submucosal ulcerated mass in the distal transverse colon.

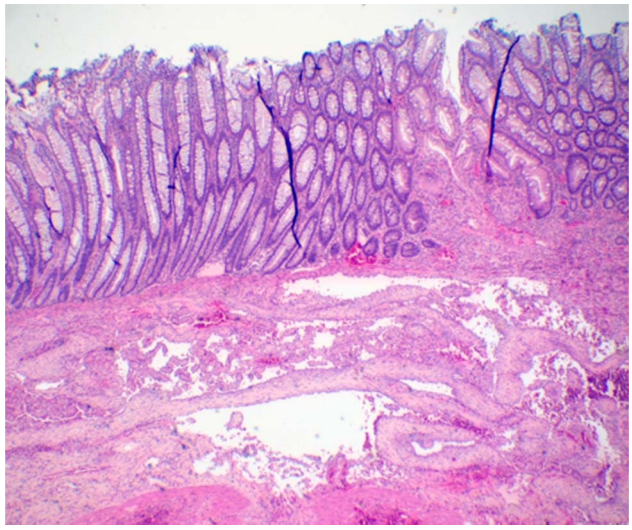

Figure 2. Plump epithelioid endothelial cells with atypical vascular proliferation (40× magnification).

Staging imaging with computed tomography (CT) thorax, abdomen, and pelvis and positron emission tomography-CT showed no evidence of metastatic disease, which confirmed the diagnosis of PCEA. One week later, segmental transverse colectomy was performed, and pathology revealed a pT2pN0Mx epithelioid angiosarcoma with clear margins. Since that time, the patient has had yearly surveillance colonoscopies and positron emission tomography-CT scans which showed no recurrence of the tumor. Three years later, he is alive and has had no further complications related to his cancer.

DISCUSSION

While angiosarcomas are an aggressive subtype of soft-tissue sarcomas that can develop anywhere in the body, primary colonic tumors are particularly rare.[1] To our knowledge, there are only 13 other reported cases of PCEA in the literature.[2–4] The symptoms of PCEA are nonspecific: most commonly rectal bleeding and abdominal pain. Endoscopically, it typically presents as an ulcerated mass which may also bleed. As with all angiosarcomas, the mainstay of treatment for PCEA is resection with negative margins; however, the prognosis is almost universally poor, with most patients dying within 1 year of diagnosis.[5]

The causes of angiosarcoma in general are not well understood, and there are several proposed risk factors including radiation/chemical exposures, genetic mutations, lymphedema, and even foreign body retained in tissue.[6] Our patient did not have any known risk factors. Histologically, epithelioid angiosarcomas consist of endothelial cells bearing an epithelioid appearance and are difficult to distinguish from both normal tissue and other types of neoplasms such as carcinomas or benign hemangiomas.[7] The positive CD31 and D2-40 stains in our patient's pathology samples helped establish the diagnosis.

Our patient's case is unique in that the cancer was found at the site of his previous polypectomy. The endoscopic history, time-course, and pathology seem to suggest that the initially visualized "polyp" was not a polyp, but in fact the angiosarcoma growing in the submucosa giving the appearance of a polyp. The removal of the overlying benign colonic mucosa allowed the tumor to bleed into the lumen. Of further note, the patient is alive and has had no recurrence 3 years after his tumor resection. The only other reported case of PCEA with survival greater than 2 years after diagnosis involves a woman treated with posterior pelvic exenteration. The report's authors hypothesize that her survival is likely due to the profound and extensive intervention she underwent.[8]

While PCEA is a rapidly progressing disease unlike the relatively indolent colorectal adenocarcinoma, its rarity and nonspecific presentation of symptoms preclude any meaningful screening strategies. Our patient's case further highlights this fact in that his presentation seemed entirely consistent with persistent postpolypectomy bleeding, although it clearly was not. In addition, our patient's cancer-free survival after segmental transverse colectomy is singularly unique in that it demonstrates that extensive surgery may not be required for successful treatment.

DISCLOSURES

Author contributions: RB is the primary author and was involved in research, analysis, and manuscript composition and is the author guarantor. CM contributed to the case and provided expertise. DM contributed pathology slides and provided expertise. JH is the senior researcher and was involved in research, analysis, and manuscript composition.

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Informed consent was obtained for this case report.

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