

Abdominal Pain in A 82-Year-Old Male: A Case Report

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ABSTRACT

Angiosarcoma is a rare malignancy which arises from the inner lining of blood vessels and lymph vessels. Patients most frequently present with cutaneous lesions. Vascular sarcomas are clinically aggressive and have a poor 5-year survival rate of approximately 20-35%. Primary gastro-intestinal angiosarcomas are extremely rare, and distant dissemination is infrequent. Diagnosis is challenging because their clinical presentation is atypical. In this short report, we present an uncommon case of an angiosarcoma located in the cecum of a 82-year-old male. The patient was treated with surgical resection, however he passed away four weeks later. This report highlights the recommended treatment, endoscopic findings and clinical symptoms.

Keywords: Abdominal Pain, Endoscopy, Cancer, Anemia

Introduction

A 82-year old man was referred for a second opinion because of a 2-year history of abdominal pain with concomitant weight loss. The patient had a prior history of cardiovascular disease, including aortic dissection, superior mesenteric artery dissection, and pacemaker implantation. At time of presentation, the patient experienced continuous abdominal pain located in the right lower quadrant, which was unrelated to oral intake. Weight loss was estimated to be sixteen kilograms in the last four months. A colonoscopy was performed revealing a dark ulcerative lesion in the cecum, depicted in figure 1. Histopathology showed colonic mucosa infiltrated by an atypical epithelioid proliferation. The lesional cells had prominent, irregular nuclei with an open-work chromatin and an amphiphilic cytoplasm. Cytokeratin AE1/AE3, CDX2, calretinin, PSA, Melan A and Sox10 stains were negative. The cells showed expression of CD31 and ERG. CD34 and D240 were negative. Some cells showed CD68 positivity. The lesion was located at the exact same place of a piecemeal resection of a sessile serrated lesion 6 months ago (pathology; low grade dysplasia). Blood results showed a normocytic anemia (4.9 mmol/L), elevated BSE (120 mm), leukocytosis ($14.12 \cdot 10^9/L$), and elevated C-reactive protein (104.5 mg/L). Additional CT-scan and PET-CT scan showed thickening of the cecal wall (FDG-avid) and two intraluminal irregular

masses in the descending aorta (non FDG-avid). There were no signs of ischemia or suspicious lymph nodes. What is most likely the diagnosis?



Figure 1: Endoscopic view of a dark ulcerative lesion in cecum with an elevated border.

Answer

The biopsy showed an angiosarcoma.

According to the ESMO-EURACAN-GENTURIS Clinical Practice Guideline the recommended treatment for localised angiosarcoma includes surgery, chemotherapy, radiotherapy or a combination. Treatment for advanced, clinically resectable angiosarcoma with isolated metastases includes surgery with optional chemotherapy (Gronchi *et al.*, 2021). In the light of patients' multi-morbidity, age and suspected toxicity of the chemotherapy, this was not an option. Also, sampling of the aortic lesion was omitted because of a lack of clinical consequence.

However, due to severe abdominal pain and a strong wish of the patient, he was referred to the surgeon for a right hemicolectomy. The pathology was consistent with a high grade angiosarcoma with invasion of the subserosa and a diameter of 4.5 cm, R0 resected. Four of 8 harvested lymph nodes contained vital tumor cells. Three days after surgery patient was discharged from the hospital. The patient passed away four weeks later. Autopsy was not performed.

Angiosarcoma is a rare malignancy which arises from the inner lining of blood vessels and lymph vessels. Patients most frequently present with cutaneous lesions (Young *et al.*, 2010). The estimated incidence of vascular sarcoma is 0.01 per 100,000 people per year in Europe (Gatta *et al.*, 2017). Schizas, *et al.* performed a systematic review in December 2020 and found 110 cases of primary gastro-intestinal angiosarcoma (Schizas *et al.*, 2022). They state that primary gastro-intestinal angiosarcomas are

extremely rare, and distant dissemination is infrequent (Allison *et al.*, 2004; Sadhu *et al.*, 2010; Sherid *et al.*, 2013; Mokutani *et al.*, 2017). Endoscopic findings of a gastro-intestinal angiosarcoma mainly include centrally ulcerated, hemorrhagic, highly erythematous or purpuric nodules/ masses (Saad *et al.*, 2013).

Learning point of this case is that an angiosarcoma can be found in the gastrointestinal tract and that it may be considered as a differential diagnosis of an ulcerative tumor seen during endoscopy.

Discussion

Diagnosis is challenging because their clinical presentation is atypical. However, complaints of patients mainly include abdominal pain and gastrointestinal bleeding (Schizas *et al.*, 2022).

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RO collected and interpreted the patient and literature data and wrote the manuscript. RB was contributor in writing the manuscript. Both authors read and approved the final manuscript.

Reference

Allison KH, Yoder BJ, Bronner MP, Goldblum JR, Rubin BP. Angiosarcoma involving the gastrointestinal tract: a series of primary and metastatic cases. *Am J Surg Pathol* 2004; 28: 298–307.

Gatta G, Capocaccia R, Botta L, Mallone S, De Angelis R, Ardanaz E, Comber H, Dimitrova N, Leinonen MK, Siesling S, van der Zwan JM. Burden and centralised treatment in Europe of rare tumours: results of RARECAREnet—a population-based study. *Lancet Oncol* 2017; 18: 1022–1039.

Gronchi A, Miah AB, Dei Tos AP, Abecassis N, Bajpai J, Bauer S, Biagini R, Bielack S, Blay JY, Bolle S, Bonvalot S. Soft tissue and visceral sarcomas: ESMO–EURACAN–GENTURIS Clinical Practice Guidelines for diagnosis, treatment and follow-up. *Ann Oncol* 2021; 32: 1348–1365.

Mokutani Y, Hata T, Miyake Y, Kuroda H, Takahashi H, Haraguchi N, Nishimura J, Mizushima T, Yamamoto H, Doki Y, Mori M. (2017) “Metastasis from a primary hepatic angiosarcoma to the colon: A case report and literature review. *Oncol Lett* 2017; 13: 2765.

Saad A, Cappell MS, Amin M. Endoscopic findings with GI angiosarcoma correspond with the propensity of these vascular tumors to cause GI bleeding: two case reports and review of the literature. *Dig Dis Sci* 2013; 58: 1797–1801.

Sadhu S, Pattari S, Shaikh F, Verma R, Roy MK. Colonic metastasis from subcutaneous angiosarcoma: A diagnostic dilemma. *Indian J Surg* 2010; 72: 328–330.

Schizas D, Mastoraki A, Giannakodimos I, Giannakodimos A, Ziogou A, Katsaros I, Frountzas M, Koutelidakis I, Vassiliu P, Pikoulis E. Primary Angiosarcoma of the Gastrointestinal Tract: A Systematic Review of the Literature. *J Invest Surg* 2022; 35: 400–408.

Sherid M, Sifuentes H, Brasky J, Shah DA, Ehrenpreis ED. Clinical and endoscopic features of angiosarcoma of the colon: Two case reports and a review of the literature. *J Gastrointest Cancer* 2013; 44: 12–21.

Young RJ, Brown NJ, Reed MW, Hughes D, Woll PJ. Angiosarcoma. *Lancet Oncol* 2010; 11: 983–991.