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Obscure gastrointestinal bleeding due to multifocal intestinal angiosarcoma

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ABSTRACT

INTRODUCTION: Intestinal angiosarcomas are an extremely rare and aggressive vascular tumors, with a few cases reported in the literature.
PRESENTATION OF CASE: A 45 years-old male arrived to our hospital with intermittent gastrointestinal bleeding presenting melena and weight loss, he has antecedent of pelvic radiotherapy ten years before admission for an unknown pelvic tumor. Emergency surgery was required because of uncontrolled bleeding and hemodynamic instability. Histopathological findings revealed a multifocal high-grade epithelioid angiosarcoma, with cells reactive for CD31, keratins CKAE 1/AE3 and factor VIII.
DISCUSSION: Angiosarcomas are aggressive tumors with a high rate of lymph node metastasis and peripheral organs. The diagnosis is difficult because it present nonspecific clinical presentation, radiological and histopathological findings. There are few reports of angiosarcoma involving the small intestine and the most common presentation are abdominal pain and gastrointestinal bleeding. There is not enough information for intestinal angiosarcoma secondary to radiation therapy, but there have been proposed criteria for diagnosis: no microscopic or clinical evidence of antecedent malignant lesion, angiosarcoma presented in the field of irradiation, long latency period between radiation and angiosarcoma and histological confirmation. We suspect our patient course with a secondary form of angiosarcoma. Therapy for bleeding angiosarcoma consists in control of bleeding and medical management to stabilize the patient. Once accomplished surgical resection is required.
CONCLUSION: We should keep in mind this tumors as a cause of obscure intestinal bleeding in patients with medical history of radiation therapy.

1. Introduction

Angiosarcomas are a very rare and aggressive tumors of vascular cell origin, they comprise 1–2% of all sarcomas. They can occur anywhere on the body but they occur mainly in the skin, soft tissue and breast and rarely the gastrointestinal tract [1,2]. They can be sporadic or secondary to some predisposing factors like radiotherapy, chronic lymphedema, familial syndromes and exposure to various chemicals. The distribution is similar between males and females and they are more common in old people [3]. These neoplasms have a very poor prognosis due to its diagnostic is usually delayed because of its diverse clinical presentation and none specific symptoms [4]. The occurrence in small intestine is extremely rare, with a few reports in the international journals [5]. We describe a case of patient who presented gastrointestinal bleeding due to multifocal intestinal angiosarcoma.

2. Clinical case

A 45 years-old male was admitted in our hospital with a 3 month history of intermittent gastrointestinal bleeding presenting melena and weight loss of 20 kg. At his arrive, he reported epigastric pain and occasional fever since a month ago. The patient referred a medical history of 20 pelvic radiotherapy sessions in a private hospital for unknown pelvic tumor 10 years before its admission. Diagnostic workup began with an upper endoscopy and colonoscopy with a negative report of visible bleeding and non-anatomical abnormalities. A computer tomographic scan of the abdomen showed irregular and circumferential thickening of jejunum, with 24 mm of thickness, without lymph nodes and other intra-abdominal lesions (Figs. 1 and 2). Serum levels of tumors markers ACE and Ca 19.9

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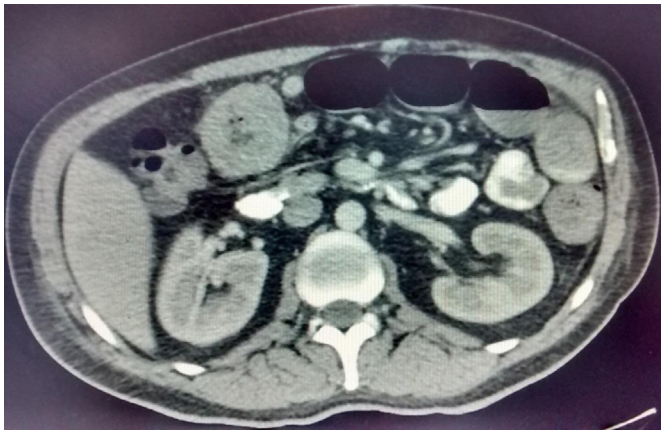

Fig. 1. CT scan, axial plane show an intestinal tumor.

were normal, except Ca 125 that was increased 4 fold normal value. Conservative management was started with hemodynamic stabilization and multiple transfusions (packed red blood cells and fresh frozen plasma).

We decided an emergency surgery in his third day of admission because high transfusional requirements and hemodinamical instability. An exploratory laparotomy was performed finding macroscopic evidence of four tumors at 15, 30, 50 and 150 cm distal to Treitz's ligament (Fig. 3); therefore, three intestinal resections with enteroenterostomy and omentectomy were performed. No transoperative complications were reported. The postoperative course was complicated by surgical site infection and the patient

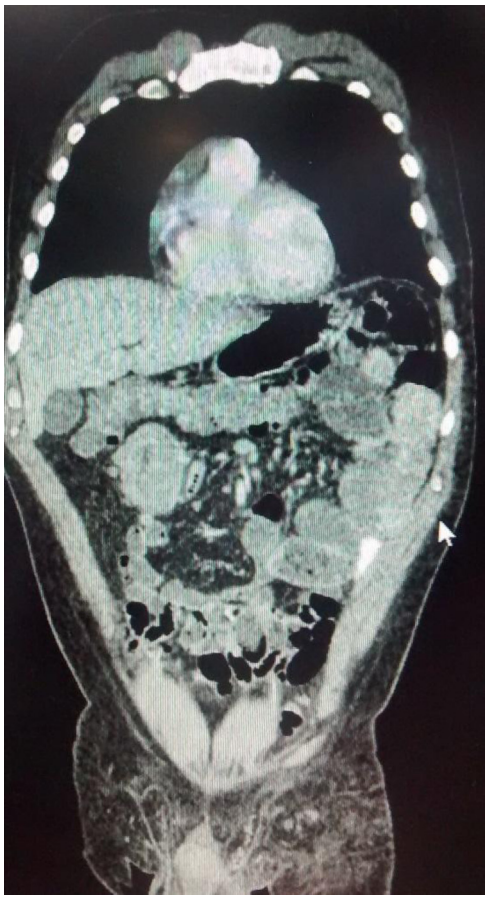

Fig. 2. CT scan, coronal plane showing thickening of intestinal wall.

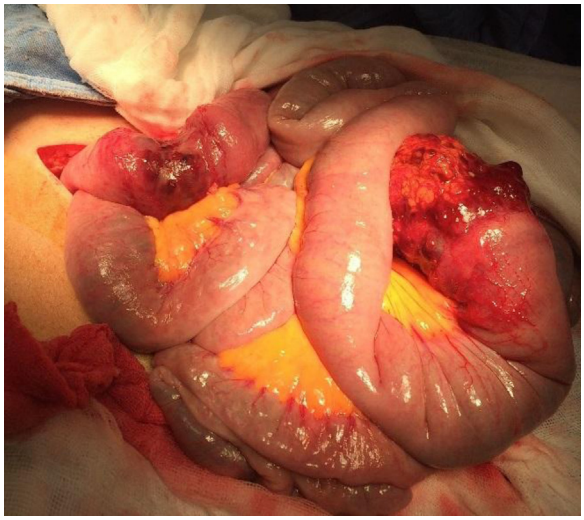

Fig. 3. Intraoperative presentation of the multifocal tumors.

was treated with IV antibiotics, he was discharged 10 days after surgery. The patient was referred to Oncology center to receive adjuvant therapy.

The pathological findings revealed a multifocal high-grade epithelioid angiosarcoma, with cells reactive for CD31, keratins CKAE 1/AE3 and factor VIII, and nonreactive for CD34 involving ileum and jejunum with diameter 4.5, 6.5 and 8.5 cm, highly vascularized, ulcerated and infiltrating tumors, with focal vascular permeation, without lymphatic and neural permeation, no necrosis. Six omentum metastatic implants were reported.

3. Discussion

Primary angiosarcoma of the small intestine is an extremely rare disease affecting mainly: liver, spleen, adrenals, ovaries, heart, lung, breast and, rarely, the middle gastrointestinal tract as in our case [2]. Angiosarcomas are aggressive tumors with a high rate of lymph node metastasis and peripheral organs. The diagnosis is difficult because of nonspecific clinical presentation, radiological and histopathological findings, so the angiosarcoma of the small intestine represents unique challenge to diagnose [6].

These tumors could have clinical presentation with bleeding of gastrointestinal tract, anemia, intestinal obstruction, abdominal pain, nausea, abdominal distention, weight loss, shortness of breath, weakness and diarrhea. In our case, present a patient with gastrointestinal bleeding like initial symptom [2,7,8]. There is a recent report presented giant abdominal angiosarcoma (diameter > 20 cm) [7].

Ni et al., reported 27 cases of angiosarcoma involving the small intestine [2]. The most common presentation was abdominal pain and gastrointestinal bleeding (37%). Twenty-three patients presented primary angiosarcoma (85.15%), 4 patients indeterminate (14.8%) and none with secondary form of angiosarcoma. Nine patients had medical history of radiation (33.3%) all of these with the primary form. One patient had multifocal intestinal of angiosarcoma and no history of radiation, being a primary form of angiosarcoma. Our case, as we have described, the patient presented gastrointestinal bleeding caused by a multifocal intestinal angiosarcoma, and because the history of pelvic radiation, we supposed that, this presentation was a secondary form of intestinal sarcoma [2].

Angiosarcomas are classified as well-differentiated, poorly differentiated, and epithelioid tumors [5]. The histopathological diagnosis is difficult because it shows high architectural and