



Case Report

Carcinosarcoma of the colon with extensive and extraordinary metastases detected on F-FDG18PET/CT: A case report

Meryem Maskrout^{a,*}, Farah Boutaggout^a, Rania Mokfi^a, Ghizlane Ennibi^b, Youssef Hnach^c, Mohamed Tarchouli^d, Ghizlane Rais^a

^a Medical Oncology Department, Souss Massa University Hospital Centre, Faculty of Medicine and Pharmacy of Agadir, Ibn Zohr University, Agadir, Morocco

^b Nuclear Medicine Department, Oncology Center of Agadir Souss Massa University Hospital Centre, Agadir, Morocco

^c Gastroenterology Department, Oued Eddahab Military Hospital, Faculty of Medicine and Pharmacy of Agadir, University Ibn Zohr Agadir, Morocco

^d General Surgery Department, Oued Eddahab Military Hospital, Faculty of Medicine and Pharmacy of Agadir, Ibn Zohr University, Agadir, Morocco



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ABSTRACT

Introduction: Sarcomatoid carcinomas or carcinosarcomas are rare tumors with a double component, carcinomatous and sarcomatous. They most commonly arise from the head, neck, respiratory system and female genital tract. To the best of our knowledge, only thirty two cases of colorectal involvement have been reported in the medical literature.

Case report: We report a case of sarcomatoid carcinoma of the colon in a 58 year old woman with unusual sites of metastasis revealed by a whole body 18F-fluoro-2-deoxyglucose positron emission tomography/computed tomography (FDG-PET/CT), who presented with right iliac fossa pain. Physical examination revealed only tenderness of the right iliac fossa. A right hemi-colectomy was performed. The immunohistochemical study of the surgical specimen revealed 2 cellular contingents, one carcinomatous and the other sarcomatous, leading to the diagnosis of carcinosarcoma. The evolution was rapidly unfavourable, with death occurring 4 months after the initial surgery.

Discussion: Colorectal carcinosarcomas are very rare and aggressive tumors with high metastatic potential commonly in the liver, lymph nodes, and peritoneum. In our case, we order a FDG-PET/CT that revealed intriguing pattern of metastasis.

Conclusion: The purpose of presenting this case report is to raise awareness among clinicians to consider this clinical entity as a differential diagnosis of colorectal tumors and order more often FDG-PET/CT for a good clinical staging.

1. Introduction

Carcinosarcomas or sarcomatoid carcinomas are biphasic tumors with exhibiting epithelial and stromal malignant differentiation [1]. They can arise from any location but occur most often in the head, neck, respiratory system and female genital tract [2]. Colorectal carcinosarcomas are extremely rare [3]. There are few reports of individual cases, but no large series from a single institution has been published. It is a very aggressive tumour often diagnosed at the metastatic stage [4]. The most common metastatic sites are the liver, lung and peritoneum [5]. Surgical treatment remains the standard of care in early colorectal carcinosarcomas as well as adenocarcinomas. We report a case of carcinosarcoma of the colon with unusual sites of metastasis revealed by

FDG-PET/CT scan. This work has been reported in line with the SCARE 2020 criteria [6].

2. Case report

We report a case of carcinosarcoma of the cecum in a 58-year-old woman with unremarkable personal and family history who presented with right iliac fossa pain that appeared 3 months before, without other symptoms, in particular no rectal bleeding, vomiting or subocclusive syndrome. Physical examination revealed only tenderness of the right iliac fossa. An abdominal ultrasound was performed revealing a caecal mass. Routine biological assessment, including liver and kidney function tests and complete blood count, was unremarkable. Abdominal and pelvic computed tomography revealed a thickening of the colonic wall

* Corresponding author. 05, Imm 65, Résidence Jardins De Souss, Hay Mohammadi, Agadir, 80000, Morocco.

E-mail address: meryem.maskrout@gmail.com (M. Maskrout).

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Abbreviation

PET/CT	Positron Emission Tomography/Computed Tomography
FDG	Fluorodeoxyglucose
SUV	standardized uptake values

at the lower cecum with two large adrenal masses of 7 cm each. With this presentation, we suspected either a colonic adenocarcinoma, a neuroendocrine tumor, a gastrointestinal stromal tumor (GIST) or a caecal carcinosarcoma.

Colonoscopy was performed and revealed a non-stenotic, highly vascularised caecal swelling process. Unfortunately, a biopsy could not be performed because the gastroenterologist judged that it was very dangerous due to the high risk of bleeding and because of a lack of logistic resources. Two days after the colonoscopy, the patient presents an acute abdomen on caecal volvulus for which she underwent emergency surgery. A right hemi colectomy was performed under general anesthesia by the General Surgery Department of a Military Hospital. Post-operatively, no complications or adverse effects have been reported.

Anatomopathological analysis of the surgical specimen revealed a morphological aspect of a malignant, undifferentiated, partially necrotic tumour proliferation, ulcerated on the surface (Fig. 1 A and B).

Immunohistochemistry (IHC) staining was performed. The tumor tested positive for the epithelial marker cytokeratin AE1-AE3 and mesenchymal marker vimentin which is supported the diagnosis of carcinosarcoma (Fig. 1C).

One month after the surgery, the patient presents pain and abdominal distension. A thoraco-abdominal and pelvic CT scan was performed showing thirty percent progression of bilateral adrenal lesions and a tumor-like formation of the aortic arch (Fig. 2).

Trans-thoracic ultrasound confirmed the presence of an irregular tissue mass after the left subclavian artery leading to a moderate stenosis. Moreover, the patient reported a painful swelling of the posterior side of the right knee. A whole body 18F-fluoro-2-deoxyglucose positron emission tomography/computed tomography (FDG-PET/CT) was applied in order to investigate additional metastatic sites and demonstrated high FDG uptake in the aortic arch (SUV max 11.8) (Fig. 3 A), two huge necrotic adrenals measuring 11 cm each (SUV max 18.12) (Fig. 3 B), two right flank peritoneal nodes (SUV max 13.9), a right ischio-pubic osteolytic mass (SUV max 19.5) (Fig. 3C), and an osteomedullary lesion of the right femur (SUV max 19.4) (Fig. 3 D).

After an Multidisciplinary Team Meeting and because of stage 4 colorectal carcinosarcoma, palliative chemotherapy with 5-fluorouracil and adriamycin was started with poor digestive tolerance. Systemic therapy was interrupted due to poor tolerance and deterioration of the patient's general condition with a WHO performance status becoming 3

to 4. Death occurred 4 months after the initial surgery.

3. Discussion

Three main reasons to present this case report. The first reason is the rarity of colorectal localization, the second one is the very unusual metastatic sites in colorectal cancers as arterial metastasis and bilateral adrenal metastasis. The third reason for reporting this case is the use of FDG-PET/CT to detect and assess all metastatic sites. However, the limitation in our case that during the colonoscopy, no biopsy was performed even if the mass appeared highly suspicious of malignancy.

Carcinosarcoma occurs mainly in the head, neck, respiratory system and female genital tract [2]. In the digestive tract, it was reported mainly in the oesophagus and stomach [2]. The first case of colorectal carcinosarcoma was reported in 1986 by Weidner et al. [7]. To the best of our knowledge, no large series has documented the exact number of colorectal carcinosarcoma's cases worldwide. Data are available in isolated case reports. To date, only thirty two cases have been reported [4].

Clinically, colorectal carcinosarcoma resembles other colorectal tumors in symptomatology, physical signs, and radiology. In a review recently reported by Kyriakos and al. [4], the clinical features of colorectal carcinosarcomas were summarized. The average age of occurrence was 64 years with extremes of age between 13 and 84 years. Rectorrhagia and abdominal pain were often suggestive of colonic tumor. The primary tumour was located mainly in the rectum in 29% of cases, while caecal location was found in only 3 patients (9%) [4]. In our case, colorectal carcinosarcoma was found in a 58 year old patient with abdominal pain. The tumor in our patient occurred in the cecum and thus considered the fourth case reported in this location. The clinical specificity of our case was the rapid evolution to occlusion upon volvulus of the cecum.

Histologically, carcinosarcoma consists of a mixture of an epithelial component most often a mild to high grade adenocarcinoma and a sarcomatous element of mesenchymal origin which may be differentiated or undifferentiated [8]. Further characterization of the nature of the tumour itself has been achieved through immunohistochemistry, confirming the presence of distinct components. The most commonly observed pattern of staining is reactivity in the adenocarcinomatous component to the epithelial markers, cytokeratin 20 (CK20) and Carcinoembryonic antigen (CEA) [5,9]. The sarcomatous cells frequently stain positively for vimentin, desmin, and SMA [1,7,9]. In our patient, the tumor tested positive for cytokeratin AE1-AE3 and vimentin.

Full assessment is required before starting treatment. The minimum requirements for staging include chest, abdominal and pelvic imaging by CT scan, which are necessary to localize the tumor accurately, and to provide information about tumor size and involvement of adjacent organs. It may help in surgical planning if indicated and investigate metastatic sites.

The FDG-PET/CT may offer the unique opportunity to provide not only whole-body images, but also metabolic and functional information

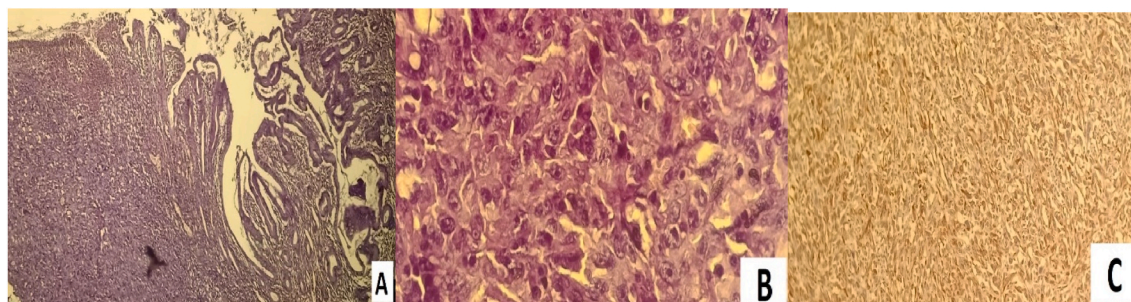


Fig. 1. anatomopathological examination of the intestinal mucosa with a diffuse and poorly differentiated tumour proliferation with a sarcomatoid appearance (HEx4) (A), the tumour cells show pronounced atypia (HEx40) (B), immunohistochemical study showing that tumour cells express cytokeratin AE1-AE3 in an intense and diffuse manner (C).

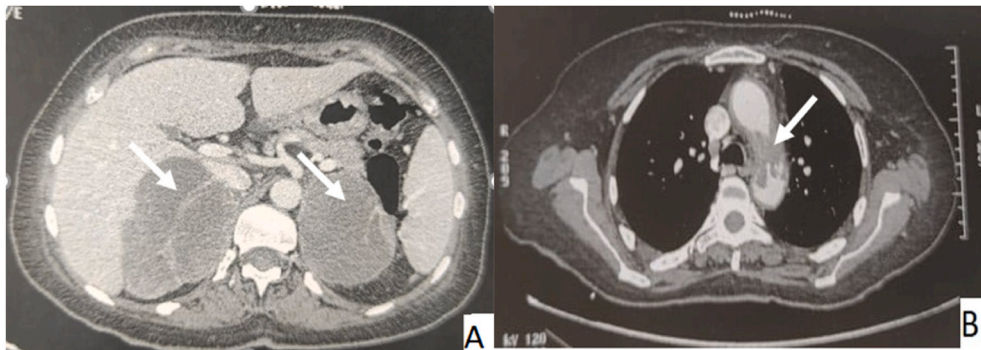


Fig. 2. Post op CT scan showing thirty percent progression of bilateral adrenal lesions (A) and a tumor-like formation of the aortic arch (B).

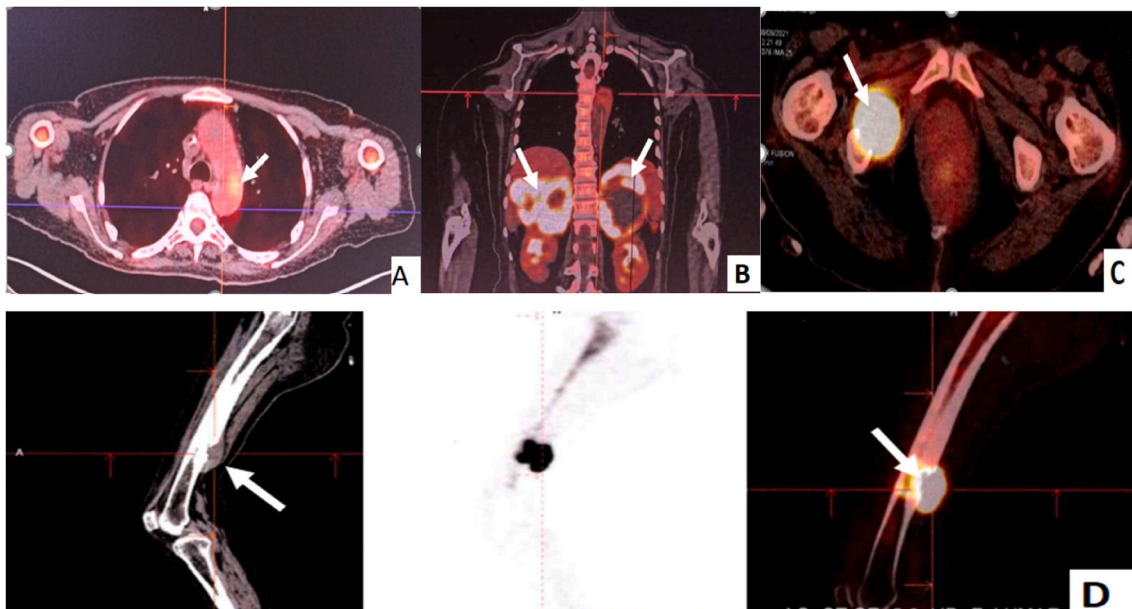


Fig. 3. Pet/CT showing high FDG uptake in the aortic arch (SUV max 11.8) (A), two huge necrotic adrenals measuring 11 cm each (SUV max 18.12) (B), a right ischio-pubic osteolytic mass (SUV max 19.5) (C) and an osteomedullary lesion of the right femur (SUV max 19.4) (D).

regarding tumor tissue. Moreover, the widespread availability of this imaging technique led to increasing the number of incidental findings [10,11]. It was of great help for a better clinical staging. In our case, it was performed in order to investigate additional metastatic sites and demonstrated high FDG uptake in the aortic arch, two huge necrotic adrenals measuring 11 cm each, two right flank peritoneal nodes, a right ischio-pubic osteolytic mass, and an osteomedullary lesion of the right femur. These metastatic sites were rarely observed in colorectal adenocarcinomas and indicate an unpredictable and aggressive evolution of the disease, mainly due to the sarcomatous components.

The prognosis of colorectal carcinosarcoma is poor mainly due to the advanced stage of the diagnosis in most cases, but also to the rapid evolution of the disease with the appearance of multiple metastases and rapid alteration of the general condition. A review of thirty two previous cases found synchronic metastatic disease in eleven patients with the most common sites being liver, lymph nodes, and peritoneum [4]. The particularity of the present case is the unusual locations of metastasis. Indeed, our patient had huge adrenal metastases, as well as secondary bone and intra aortic lesions. A mesenteric mass at the aortic bifurcation has been reported in the literature [7]. Bone and adrenal locations have never been described.

The median overall survival of patients was few months which confirms the poor prognosis of colorectal carcinosarcomas. Only 4 patients were alive 2 years after the initial surgery [4] although Susumu

Ohwada et al. reported a case of 18 years disease-free survival for carcinosarcoma of the transverse colon [12].

Since only a few cases have been reported and the disease is rapidly progressing, the therapeutic management remains a challenge. In all cases including our patient, patients had surgical resection of the primary colorectal tumor, whereas only 13 patients (40%) were candidate to chemotherapy [4]. The rarity of colorectal carcinosarcomas mostly reported as isolated cases did not allow to define the optimal medical treatment. Several chemotherapy protocols have been used mostly based on 5 fluoro uracil: 5FU-mitomycin, endoxan-doxorubicin-cisplatin, 5FU-leucovorin, or gemcitabine-taxotere [4]. Antiangiogenic drugs such as Bevacizumab was associated to chemotherapy in one case [13]. In our case, after an Multidisciplinary Team Meeting, we decide to use palliative chemotherapy based on 5-fluorouracil and adriamycin.

4. Conclusion

The purpose of presenting this case report is not only to report a colorectal carcinosarcoma with uncommon metastasis sites and fatal evolution but also to raise awareness among clinicians to add FDG PET/CT as a potential modality for diagnosing advanced disease by providing whole-body cancer involvement information (i.e., N and M staging) that might not be clinically apparent.

Ethical approval

This is a case report that does not require a formal ethical committee approval.

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The authors declared that this study has received no financial support.

Author contribution

Meryem Maskrout and Ghizlane Rais were involved in the analysis of the data and the literature search and wrote the manuscript. Farah Boutaggout, Ennibi Ghizlane, Mohamed Tarchouli and Youssef Hnach helped with the patient management and revision of the manuscript. Rania Mokfi contributed to the preparation of this manuscript, and interpretation of the case. All the authors have read and approve the final version of the manuscript.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Registration of research studies

1. Name of the registry:
2. Unique Identifying number or registration ID:
3. Hyperlink to your specific registration (must be publicly accessible and will be checked):

Guarantor

Meryem Maskrout.

Provenance and peer review

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Declaration of competing interest

The authors declare no conflict of interest.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.amsu.2022.104450>.

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