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Endometrial Stromal Sarcoma as a Second Primary in a Patient with Colorectal Carcinoma: Review of the Literature

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ABSTRACT

We are presenting here the first case of second primary endometrial stromal sarcoma in a patient with colorectal carcinoma and reviewed the literatur regarding metastasis patterns in colorectal carcinomas and the managment of endometrial stromal sarcomas. The differential diagnosis of colon cancer metastasis and second primary was of great importance in our case because the management and prognosis would differ fundamentally. The presumption that metastatic cancers are incureable, precludes the work-up for detection of secondary cancers in most cases. This may hinder the proper treatment of patients. That is why the detection of secondary cancers should be strongly encouraged especially in cases with an unexpected metastasis pattern.

Key words: colorectal cancer, endometrial stromal sarcoma, second primary

KOLOREKTAL KANSER SEYRİ SIRASINDA İKİNCİ PRİMER OLARAK GELİŞEN ENDOMETRİAL STROMAL SARKOMA OLGUSU: LİTERATÜRÜN GÖZDEN GEÇİRİLMESİ

ÖZET

Literatürde kolorektal kanser seyri sırasında ilk defa saptanan endometrial stromal sarkom olgusunu sunuyoruz. Olgumuzda kolon kanseri metastazı ve ikinci primer arasında ayırım yapabilmek çok önemliydi, çünkü prognoz bilgileri ve tedavi yöntemleri tamamen değişmekteydi. Metastatik kanserin şifa olmayacağı önyargısı, çoğu zaman ikinci pirimer çalışmalarının yapılmasını ve hastaların doğru tedavilere ulaşmasını engellemektedir. Bu nedenle özellikle alışılmadık metastaz tablolarında ikinci primer araştırmaları mutlaka yapılmalı ve karar verilene kadar ısrarla tekrarlanmalıdır.

Anahtar sözcükler: kolorektal kanser, endometriyal stromal sarkom, ikinci primer

A 52 years old woman was diagnosed with pT3N1M0 colorectal adenocarcinoma after a hemicolectomy. Postoperative PET/CT evaluation revealed several abdominal lymph nodes measuring about 1 cm in diameter, with an SUV value of 5.7 and a mass inher uterus consistent with myoma uteri. She was under follow-up care for her myoma uteri for the past seven years. She was started on adjuvant chemotherapy with FOLFOX with close follow-up for these suspicious lymph nodes. Interim PET/CT evaluation after three months revealed no other pathologic finding besides her known mass in the uterus. Her adjuvant chemotherapy was completed. The last PET/CT evaluation in March 2013 revealed multiple mediastinal and intrabdominal lymph nodes with an SUV of 4.4-6.5 and a mass in the right iliac bone with a diameter of 27.5 mm and an SUV of 14.6.

The case was discussed at the tumor board. This clinical picture suggested a second primary cancer, so an interventional radiologist was requested for a biopsy of the new lesions. The location of the lymph nodes precluded their sampling. So a trucut bone biopsy was performed first which revealed an inflamatory reaction. An abdominal MRI was performed to see the uterine mass better. The lobulated mass was hypointense in the T1 series and hyper-intense the T2 series and its radio-contrast uptake pattern suggested a sarcoma. In Mai 2014 a total hysterectomy, bilateral salphingo-oopherectomy and abdominal

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lymph node sampling were performed. The pathologic examination revealed a low-grade endometrial stromal sarcoma (Desmin, SMA, Pan-CK, S100, CD34, CD117, EMA, ER, PR negativE., CD10 ve Cyclin D1 positive; Ki67 index: %25; no tumor necrosis, mitosis <10/10HPF; T:13 cm).

Because of a lack of estrogen and progesteron positivity in her tumor specimen, hormonal therapy was not done. She experienced severe coughing due to a lung nodule above the diaphram so she was started on palliative chemotherapy for her metastatic sarcoma consisting of doxorubicine and ifosphamide. She did not respond well to two cycles of chemotherapy.

Discussion

Colorectal carcinoma remains the third most common cancer among adult men and women and is the third most common cause of death from cancer (1). It is well recognized that colorectal cancer does not frequently metastasize to bone. The incidence of bone metastasis in colerectal cancer is reported to be about 10%. The most common sites of metastases are the liver and the lungs. Solitary bone metastases and bone metastasis at presentation are very uncommon (2). Liver metastases preceede lung metastases in most cases. A retrospective study evaluated 252 patients regarding the metastasis pattern in colorectal carcinoma. Analysis of metastasis to bone showed that 14 of the 252 individuals (5.5%) had bone lesions and no individuals had metastasis only to bone at the time of diagnosis. In all patients that developed bone metastasis, liver and/or lung metastasis was always present first. One individual presented with bone lesions at the time of diagnosis; however, liver metastasis was also present (3).

Endometrial stromal sarcomas (ESS) are rare malignant tumors of the uterus, and most of the information available in literature is based on small series or case reports (4). Although the main tumor mass is almost always intramyometrial, most ESS involve the endometrium and uterine curettage which may be helpful in preoperative diagnosis(5, 6). However, when the lesion is completely within the myo-metrium, the scrapings may not be helpful. Due to the great similarity between ESS and normal endometrium, it may be impossible to diagnose it with certainty on curettage fragments, and the definitive diagnosis can be made only on a hysterectomy specimen.

Strong and/or diffuse positivity for CD10 is found in ESS, which is helpful in distinguishing these tumors from histological mimics like cellular leiomyoma, that are generally negative (7). ESS is positive for both estrogen and progesterone receptors in most cases.

As for other sarcomas, surgery is the most effective treatment for ESS. The efficacy of adjuvant therapy is not proven. Survival in patients with undifferentiated endometrial sarcoma (previously called high-grade endometrial stromal sarcoma) appears to be related to the extent of residual disease after initial surgery and would suggest the necessity for aggressive cytoreduction as a main modality of treatment. However, the role of debulking surgery for ESS (formally known as "low-grade ESS") remains unclear (8). Recurrent ESS has been treated with hormone therapy, radiation, surgical re-excision, or a combination of two or more of these modalities (9). There are few case reports where the recurrent ESS was treated wit hetoposide, cyclophosphamide, and doxorubicin (10). Even though chemotherapy is a mode of treatment in undifferentiated endometrial sarcoma, data supporting their efficiency in the case of recurrence of ESS are limited. Because of the large variation in pathologic characteristics, combined with a scarcity of patients, there is insufficient information about optimal management.

Conclusion

We presented here the first case of second primary endometrial stromal sarcoma in a patient with colorectal carcinoma. In our case, the differential diagnosis of colon cancer metastasis and second primary cancer was of great importance because the treatment and prognosis is different. The preassumption that metastatic cancers are incurable precudes the work-up for detection of secondary cancers in most cases. This may hinder the proper treatment of patients. That is why the detection of secondary cancers should be strongly encouraged especially in cases with an unexpected metastasis pattern.

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