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Case Report

Radiotherapy-induced ovarian sarcoma following the treatment of cervical cancer: Exceptional case report and review of the literature

Malek Mohamed Ayadi[1], Olfa El Amine El Hadj[2], Ines Zemni[1], Hatem Bouzaiene[2], Jamel Ben Hassouna[1], Amor Gamoudi[2], Khaled Rahal[1]

[1]Department of Surgery, Salah Azaiez Institute, Tunis, Tunisia, [2]Department of Pathology, Salah Azaiez Institut, Tunis, Tunisia

Address for correspondence:
Malek Mohamed Ayadi, Department of Surgery, Salah Azaiez Institute, Tunis, Tunisia.
E-mail: melek.ayadi@gmail.com

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ABSTRACT

Primary ovarian sarcomas are rare gynecological tumors accounting for <3% of all ovarian tumors. Radiation-induced sarcoma is a rare and a serious complication of radiotherapy. We report the case of a 70-year-old patient who was treated for radiation-induced sarcoma of the ovary. To the best of our knowledge, this is the first case reported. Through this work, we will try to report an exceptional case for literature and report our experience in the treatment and the evolution of this particularly pathology.

KEY WORDS: Pathology, radio-induced, ovarian, sarcoma

INTRODUCTION

Primary ovarian sarcomas are rare gynecological tumors accounting for <3% of all ovarian tumors. Some of the largest series have found five primary sarcomas out of 3890 ovarian neoplasms. Radiation-induced sarcoma is a rare and a serious complication of radiotherapy, only one case was reported in literature [1].

CASE REPORT

We report the case of a 70-year-old patient who was treated for well-differentiated squamous cell carcinoma of the cervix. The tumor was classified as Stage IIb, treated exclusively with radiotherapy at a dose of 50 gray. The clinical response was good.

About 18 years after the patient was explored for abdominopelvic mass in the right side; the scanner showed a large ovarian formation of 120 mm diameter with a double component cystic and tissue involving the rectum. The patient had a right oophorectomy, rectal resection, and colostomy.

Histological examination showed that it is a malignant mesenchymal proliferation. Grossly, the tumor was well circumscribed, firm with hemorrhagic areas. Microscopic findings demonstrated densely cellular tumor arranged in long fascicles that are angled in a chevron-like [Figure 1]. The cells have tapered darkly staining nuclei with variable prominent nucleoli and scanty cytoplasm [Figure 2]. Increased mitotic activity was observed with 7/10 high-power fields. The tumor has a variable collagen, from a delicate intercellular network to diffuse sclerosis. Immunohistochemical study showed a diffuse expression of Vimentin [Figure 3] and CD 34 antibodies. Tumor cells were actin smooth muscle and PS 100 negatif.

After 2 months, she had ovarian staging without lymphadenectomy. The patient was followed for 8 years with no signs of local or distant recurrence.

DISCUSSION

The cervical cancer is a curable disease often with a survival rate of nearly 85% in Stages I and II and more than 50% in case of advanced tumor.

The locoregional control rate has improved in recent years thanks to the chemo-radiotherapy and brachytherapy.

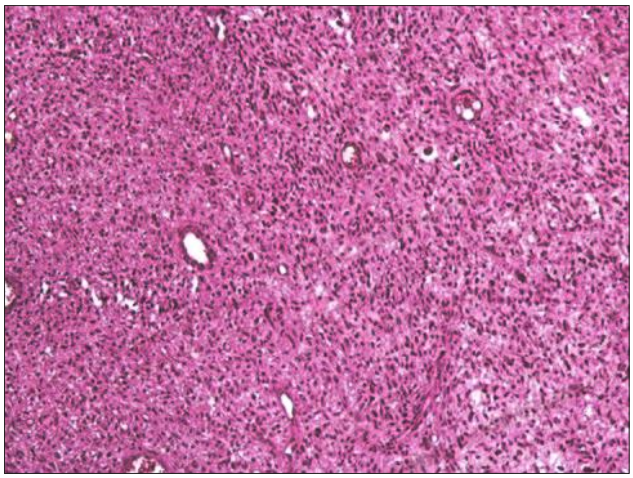

Figure 1: Densely cellular tumor arranged in long fascicles that are angled in a chevron-like

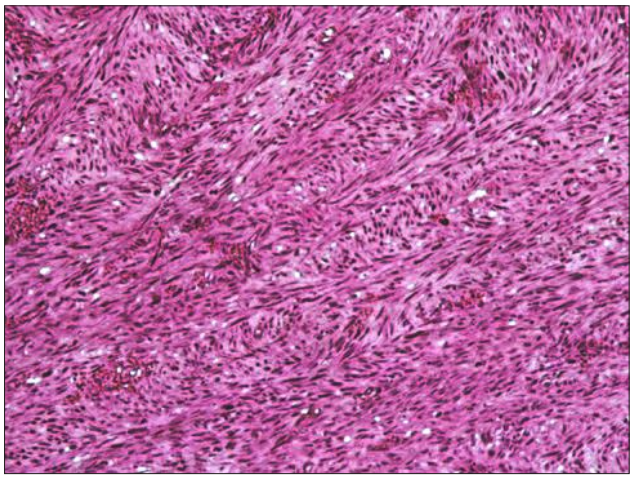

Figure 2: Tumor cells have tapered darkly staining nuclei with variable prominent nucleoli and scanty cytoplasm. And increased mitotic activity

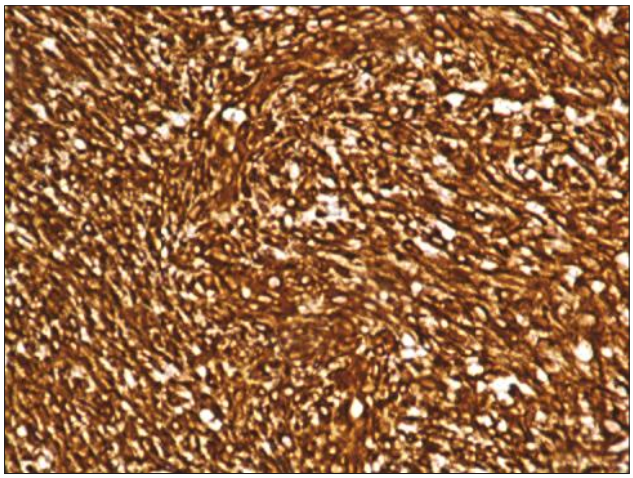

Figure 3: Immunohistochemical study showed a diffuse expression of vimentin

Radiation-induced sarcomas are a rare complication of radiotherapy. The first case of a radio-induced tumor was described in 1922 by Beck after a radiotherapy for breast carcinoma [2].

Cahan *et al*. have proposed four criteria for the diagnosis of radio-induced sarcomas: Antecedent of radiotherapy; clinical latency of several years (more than 5 years); the occurrence of Kaposi sarcoma in the irradiated field; histological confirmation of the nature of the sarcomatous post-radiation lesion.

These criteria have been later extended by Arlen *et al*., in 1971, a latency of 3 years or more and a localized tumor in irradiated or adjacent to the radiation fields [3].

Several risk factors for developing sarcomas in irradiated tissue have been described in the literature as the dose (beyond 50 Gy) [4-6]. The addition of chemotherapy in concomitant or sequential protocols can develop second cancer [7]. More irradiation is made at a young age, higher is the risk of developing radiation-induced sarcoma. Kleinerman *et al*. reported in their series that the relative risk (RR) of second cancer after treatment of cervical cancer with radiotherapy was 1.2 for the first 9 years, 1.3 for 10-19 years after irradiation and 1.6 for more than 30 years. As well, in the Japanese series presented by Ohno *et al*. the RR for the entire population was 1.2-6.7% for the first 10 years and 15.6% for 20 years and 23.8% for 30 years [8-10]. The presence of a genetic predisposition appears to be related to a higher risk of sarcoma in irradiated patients such as Recklinghausen disease, retinoblastoma, ataxia-telangiectasia syndrome, and Li-Fraumeni syndrome [11].

According to Ohno *et al*. study, tobacco increased the risk of developing second cancer with an RR of 1.4 for non-smoking patients and 1.6 for smokers [10].

Sarcomas of the ovary comprise <1% of ovarian neoplasms. Most mesenchymal lesions reported in the literature are benign. Primary ovarian fibrosarcomas are malignant spindle cell tumors which usually occurring in postmenopausal women. They typically present as a large, solitary, lobular, soft fleshy solid mass with hemorrhage and cystic degeneration.

The histological criteria for primary uterine fibrosarcoma have been applied including densely cellular tumor arranged in long fascicles that are angled in a chevron-like, nuclear atypia, pleomorphism, and high mitotic rate with atypical mitosis. FIGO staging and therapy of ovarian sarcomas have traditionally been the same as for epithelial ovarian carcinomas [12].

The differential diagnosis of ovarian fibrosarcoma includes leiomyosarcoma, fibrothecoma, endometrial stromal sarcomas, mixed Müllerian tumor, spindle cell carcinoma, metastatic gastrointestinal stromal tumor (GIST), and metastasis.

The gold standard treatment of sarcomas remains surgery, in fact, the treatment consists in a wide resection of the tumor,

but unfortunately the number of operable patients is limited to less than 50% since these tumors are often diagnosed at an advanced stage.

Chemotherapy has been proposed for some histological types of radiation-induced sarcomas, Arnaout *et al*. proposed a chemotherapy treatment of radiation-induced breast cancer [13] and Bacci *et al*. for radiation-induced osteosarcomas benefit but was not significant [14].

The prognosis of radiation-induced sarcomas remains dark with a less survival rate than other sarcomas [5,15-17].

Obviously, survival depends on the histological type of sarcoma [18].

CONCLUSION

Ovarian sarcoma is a rare disease and radio-induced sarcoma of the ovary is even rarer [19]. The case reported shows a very important local invasion that characterize this type of sarcoma but no metastases, surgery made of hysterectomy bi oophorectomy without lymph node dissection has been successful a cure with no recurrence for 8 years.

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