Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Radiation Oncology

Radiation-Induced Uterine Stromal Sarcoma: A Case Report and Review of the Literature

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DOI: 10.36347/sjmcr.2024.v12i01.025 | **Received:** 16.12.2023 | **Accepted:** 22.01.2024 | **Published:** 25.01.2024

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Abstract **Case Report**

The radio-induced stromal sarcoma (RISS) is a rare clinical entity, being the second most common type of uterine mesenchymal neoplasm after leiomyosarcoma. Its incidence increases as survival after radiotherapy improves, and it often poses a therapeutic challenge. We report the case of a 58-year-old patient, followed since 2014 for initially classified stage IIb squamous cell carcinoma of the uterine cervix, treated with external pelvic radiotherapy and brachytherapy. After 6 years of surveillance, the patient presented with persistent pelvic pain. Clinical examination and pelvic MRI revealed the presence of a tumoral process in the uterine cervix with suspicious thickening of the endometrium. Morphological and immunohistochemical pathological study of the lesion confirmed its tumoral nature, indicative of a high-grade stromal sarcoma. Radiotherapy can induce malignant tumors after a latent period of several years. Radio-induced gynecological sarcomas are most often associated with a poor prognosis, emphasizing the need for careful and close monitoring after pelvic irradiation.

Keywords: leiomyosarcoma, radio-induced stromal sarcoma, radiotherapy, pelvic pain, uterine cervix.

INTRODUCTION

Radio-induced soft tissue sarcomas (RISS) are rare clinical entities. Their incidence increases as survival after radiotherapy improves, and they often present a therapeutic challenge. According to the World Health Organization (WHO) tumor classification system of 2014, endometrial stromal tumors are rare and classified into four categories: endometrial stromal nodule, low-grade endometrial stromal sarcoma, high-grade endometrial stromal sarcoma, and undifferentiated uterine sarcoma [1].

High-grade RISS is a rare tumor, especially among young women [2]. Here, we report a case of a radio-induced high-grade stromal sarcoma in a woman who presented with a pelvic mass and pain.

OBSERVATION

Mrs. S.N, a 58-year-old woman, has been under observation since 2014 for initially classified stage IIb squamous cell carcinoma of the uterine cervix. She was treated with concurrent radiochemotherapy, consisting of 46 Gy of radiotherapy along with 3 cycles of concomitant carboplatin, followed by uterovaginal brachytherapy consisting of 4 sessions of 7 Gy with good tolerance. Post-treatment evaluation did not reveal any residual lesions.

In February 2020, the patient presented to the clinic with persistent pelvic pain that had been evolving for 1 month. Pelvic MRI revealed the presence of an ulcerative and exophytic tumoral process measuring 35x45x60mm, involving the entire cervix and the upper 1/3 of the vagina. There was suspicious thickening of the endometrium with significant fluid retention and parametrial infiltration, extending to the bladder base, classified as stage IVA according to the FIGO 2018 classification. (Figure 1)

Morphological and immunohistochemical pathological examination of the lesion confirmed its tumoral nature, indicative of a high-grade stromal sarcoma. The extension assessment, including a CT scan of the chest, abdomen, and pelvis, did not identify secondary locations. The diagnosis of a radio-induced uterine stromal sarcoma was established based on the delayed onset, occurrence within the initial irradiation field, and the histological type.

Due to the impossibility of initial surgical excision, a treatment plan involving chemotherapy with doxorubicin and ifosfamide was initiated.

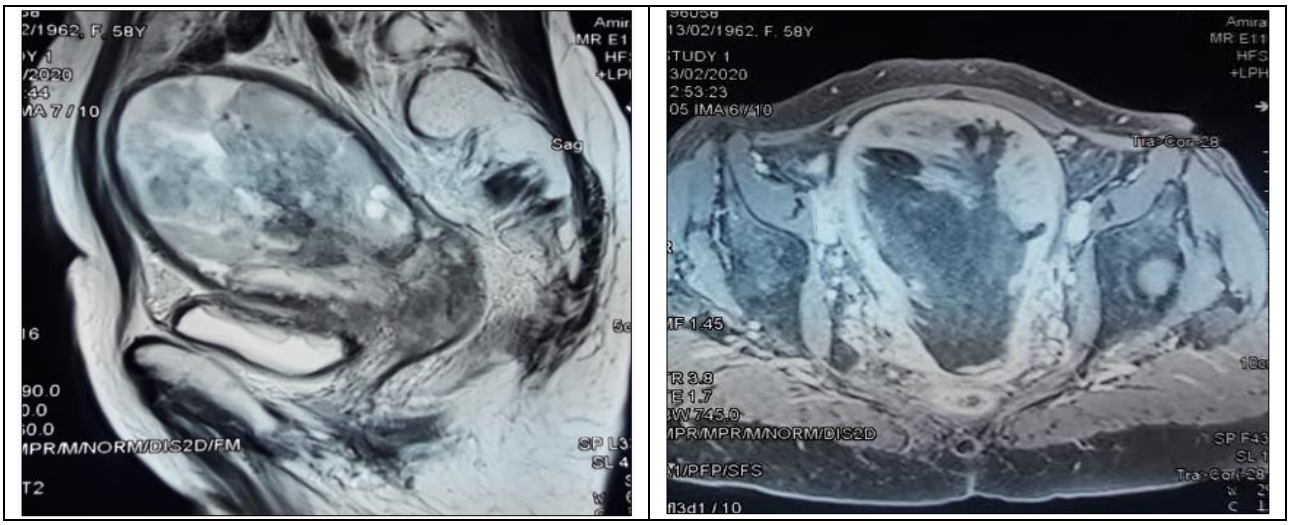

Figure 1 : Sagittal T2 and axial T1 sections of a pelvic MRI showing irregular endometrial thickening with significant fluid retention in our patient

DISCUSSION

Radio-induced sarcoma (RIS) is a well-known therapeutic complication that was initially described in the early 1900s by Perthes *et al*., Later, Cahan *et al*., introduced diagnostic criteria for RIS as histologically different lesions located within the radiation field after a latency period of more than 4 years [3]. In 2010, the Sarcoma Group at Memorial Sloan Kettering Cancer Center (MSKCC) modified this definition, stating that RIS can occur as early as 6 months after radiotherapy [4].

Gynecological radio-induced sarcoma is a rare late event after pelvic radiotherapy, with a reported incidence of 0.03% to 0.2% at 5 years and up to 0.8% overall [5]. It is considered an aggressive high-grade tumor with a poor prognosis compared to sporadic sarcoma [6].

The most common symptoms of high-grade endometrial stromal sarcoma (HG-ESS) include abnormal vaginal bleeding, palpable masses, and pelvic pain [3].

As diagnostic accuracy improves, along with the precision and high technology of radiotherapy, the incidence of sarcomas is expected to increase, including the number of radio-induced sarcomas across all locations [2].

Identifying predisposing factors for radio-induced stromal sarcoma (RISS) is a major concern. A higher incidence has been observed in patients irradiated at a younger age [7]. A multiplicative effect of radiotherapy and chemotherapy, along with a possible genetic predisposition, is under study to distinguish high-risk patients before radiotherapy [8].

Several management challenges arise due to the occurrence in a previously treated area. The standard treatment is surgery with R0 resectability, but it is less achievable than in sporadic sarcomas [9]. Previous irradiation can alter anatomical and tumor planes, hindering surgeons from accurately assessing true tumor margins [2].

Surgery may not be feasible, and systemic chemotherapy may be proposed, potentially facilitating resection. Chemotherapy includes compounds used in other soft tissue sarcomas, with anthracyclines with or without ifosfamide or dacarbazine recommended as first-line treatment in advanced diseases. The gemcitabine and docetaxel combination, trabectedin, ifosfamide, and dacarbazine are possible options for subsequent lines of treatment [10].

Adjuvant re-irradiation, if indicated, must be administered cautiously due to the higher risk of toxicities that can hinder optimal management of radio-induced sarcoma. Adjuvant radiotherapy is known to reduce local recurrence of sarcomas after conservative surgery [11]. However, as shown in Kyung Su Kim's study, due to the rarity and histological diversity within radio-induced sarcomas, analyzing the effectiveness of secondary adjuvant RT after curative resection of SSRI is challenging. Extrapolating from sporadic sarcomas, adjuvant radiotherapy should also be performed for RISS, but precautions are necessary during the patient consultation process, RT planning, and dose prescription. Additionally, high-precision RT techniques with increased conformation should be used in such re-irradiation cases to minimize toxicity [11].

CONCLUSION

The incidence of radio-induced sarcomas is on the rise. Close and vigilant monitoring after pelvic irradiation is essential. Further studies should be conducted to establish diagnostic and management guidelines for this type of cancer.

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