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Taiwanese Journal of Obstetrics & Gynecology 51 (2012) 89–92

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Case Report

Uterine carcinosarcoma associated with pelvic radiotherapy for sacral chordoma: A case report

Korhan Kahraman [a,*], Fırat Ortac [a], Duygu Kankaya [b], Gulsah Aynaoglu [a]

[a] *Department of Obstetrics and Gynecology, Ankara University School of Medicine, Ankara, Turkey*
[b] *Department of Pathology, Ankara University School of Medicine, Ankara, Turkey*

Accepted 28 December 2010

Abstract

Objective: Postirradiation sarcoma of the female genital tract is rare, but a recognized event. Most reported cases have been associated with history of radiotherapy for various gynecologic conditions, particularly cancer of the uterine cervix and abnormal uterine bleeding. The occurrence of uterine sarcoma secondary to radiotherapy for a non-gynecologic tumor and, furthermore, this condition being simultaneous with the recurrence of primary tumor is unique.
Case Report: A 67-year-old woman presented with a uterine mass which was diagnosed as a sarcoma by endometrial curettage and history of pelvic radiotherapy 23 years previously for sacral chordoma. Surgical staging procedure for uterine malignancy was performed. The final pathologic diagnosis was carcinosarcoma of the uterus.
Conclusion: In uterine masses seen in patients with history of irradiation to the pelvic field, the probability of uterine sarcomas should always be kept in mind. These tumors may occur simultaneously with recurrence of primary tumor previously treated by adjuvant radiation therapy.

Keywords: carcinosarcoma; radiation-associated neoplasm; sacral chordoma; uterine sarcoma

Introduction

Carcinosarcoma of the uterus, also referred to as malignant mixed Müllerian tumor, is a rare neoplasm composed of epithelial and mesenchymal malignant elements. These tumors accounting for 40–50% of all uterine sarcomas usually occur in postmenopausal women with a peak incidence at the ages of 60–70 years [1,2].

Previous pelvic irradiation is a recognized predisposing factor for uterine carcinosarcoma which has been estimated to occur in approximately 15% of cases [3]. Despite this defined relationship, no specific clinical or histopathological features distinguish between radiation-associated and *de novo* uterine sarcomas.

Chordoma is a rare, slowly growing, locally aggressive, malignant bone tumor that originates from notochordal remnants and occurs exclusively in the axial skeleton. The sacrum is the most common location of the chordomas [4].

Herein we present a unique case of uterine carcinosarcoma and recurrent sacral chordoma occurring simultaneously in a 67-year-old woman who had a history of pelvic irradiation for sacral chordoma 23 years previously. The clinical, radiographic, and pathologic features of this case are discussed.

Case report

A 67-year-old, multiparous woman was referred to our clinic with the diagnosis of uterine sarcoma in November 2009. Eleven days before she was referred, an endometrial curettage was performed because of postmenopausal uterine bleeding and revealed a sarcomatous malignancy at another center, but differential diagnosis between high-grade leiomyosarcoma and carcinosarcoma could not be done. She was

* Corresponding author. Department of Obstetrics and Gynecology, Ankara University School of Medicine, 06100 Ankara, Turkey.
E-mail address: korhankahraman@hotmail.com (K. Kahraman).

1028-4559/$ - see front matter
doi:10.1016/j.tjog.2012.01.018

operated because of sacral chordoma in 1986 with follow-up radiation therapy [posterior and anterior pelvic fields of 13×17 cm delivering a dose of 5000 rad (50 Gy) in 25 fractions between March and May 1986]. In addition, repeated surgeries were performed for the recurrence of sacral chordoma in 1989, 1999, and 2002. She had been menopausal since the age of 44 years and had never used hormone replacement therapy. She had no history of tamoxifen use. Her medical history was significant for diabetes mellitus and hypertension. In April 2009, a magnetic resonance imaging (MRI) scan was performed at another hospital because of a painful swelling in the lumbo-sacral region, and showed a mass indicative of sacral chordoma recurrence (Fig. 1A, B). The same MRI scan also showed a uterine mass measuring $62 \times 42 \times 38$ mm that completely filled the uterine cavity (Fig. 1A, B). Despite these MRI findings suggesting a uterine tumor, further diagnostic interventions to demonstrate or rule out uterine malignancy had not been performed at the same center. With a possible diagnosis of recurrent sacral chordoma, she had been treated with radiotherapy according to the following schedule: a total of 2600 cGy (26 Gy) in 13 fractions between April and May 2009. Follow-up MRIs did not show a significant reduction in the size of the sacral mass.

Tumor markers were normal except mildly elevated serum cancer antigen 125: 71.6 U/mL (reference range <35 U/mL). A chest radiograph and computerized tomographic scan of thorax and upper abdomen were negative for metastatic disease.

With a diagnosis of uterine sarcoma, exploratory laparotomy was performed at our clinic. Laparotomy revealed an enlarged uterus totally with a mass probably representing sarcoma. The uterine tumor invaded the bladder and the uterus was adherent to the recto-sigmoid colon, omentum, and peripheral structures. Minimal serous-hemorrhagic ascites was observed. Both ovaries were atrophic. Other pelvic and abdominal structures and peritoneal surfaces were all grossly normal. A staging procedure was performed including a peritoneal washing for cytological evaluation, total hysterectomy, bilateral salpingo-oophorectomy, para-aortic lymph node dissection, infracolic omentectomy, multiple peritoneal biopsies, and partial resection of the bladder invaded by the tumor. Pelvic lymphadenectomy was not performed because of technical difficulties resulting from the excessive fibrosis at the pelvic lymph node regions secondary to previous radiotherapy.

Macroscopic evaluation showed a polypoid mass filling the uterine cavity, 7 cm long in diameter, with a hemorrhagic, necrotic, fleshy cut surface. Histopathological evaluation of the uterine mass revealed a neoplasm composed of an admixture of malignant epithelial and mesenchymal components. The epithelial component was endometrioid type adenocarcinoma (Fig. 2A, B) and the sarcomatous areas were poorly differentiated with highly atypical spindle cells and numerous atypical mitotic figures (Fig. 2C). Areas of osteosarcoma were detected as heterologous element (Fig. 2D). Vimentin expression was strong and extensive in sarcomatous areas and focally in epithelial component (Fig. 3A). Epithelial component showed diffuse and strong cytokeratin expression immunohistochemically (Fig. 3B). Scattered smooth muscle actin (SMA) expression was detected in a few cells of sarcomatous areas indicating poorly differentiated leiomyosarcoma (Fig. 3C). There was extensive myometrial invasion exceeding the inner half. Bilateral paraovarian tissues and urinary bladder were infiltrated. Lymph node involvement was confined to only one (1/12) para-aortic lymph node. Omentum and both ovaries showed no evidence of malignancy. The cytology of peritoneal fluid was negative for malignant cells. Therefore, the final diagnosis was a FIGO (*Federation Internationale de Gynecologie et d'Obstetrique*) stage IVA uterine carcinosarcoma. She recovered uneventfully and was discharged on the 13th postoperative day in a stable condition. She received adjuvant chemotherapy consisting of six cycles of paclitaxel (80 mg/m^2) and carboplatin (2 AUC). The first three cycles were given at

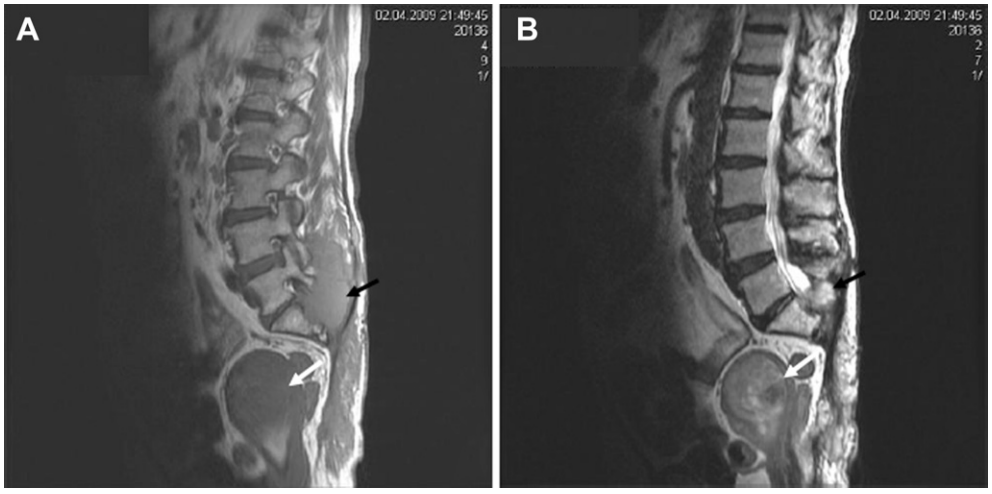

Fig. 1. (A) T1-weighted sagittal image; a hypointense recurrent sacral chordoma (black arrow) and a heterogeneous hypointense mass filling the endometrial cavity of the uterus (white arrows). (B) T2-weighted sagittal image; the relatively hyperintense uterine mass (white arrows). Hypointense areas within the endocavitary mass denote hemorrhage. The sacral chordoma (black arrow) is also seen as a relatively hyperintense mass partially filling the spinal canal and abutting the filum terminale.