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Isolated metastases in lumbar vertebrae - an extremely rare presentation of relapsed cancer of the vagina. A case report and review of the literature

Metastasi isolata in colonna lombare - una presentazione estremamente rara di cancro recidivante della vagina. Un caso clinico e revisione della letteratura

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Summary

We describe the case of a 71-year-old patient with primary squamous-cell carcinoma of the vagina (grade 3, FIGO stage I). She was treated with tumor resection followed by radiotherapy. Four months after treatment she was diagnosed with isolated metastases in the lumbar vertebrae. No synchronous abdominopelvic and/or other organ metastases were found. The patient received palliative radiotherapy to the lumbar spine. She died 17 months after primary diagnosis of cancer due to progression of the disease. We conclude that patients presenting a history of primary cancer of the vagina with chronic pain in the vertebral column should be carefully evaluated, even if the cancer is diagnosed at an early clinical stage. Eur. J. Oncol., 17 (3), 149-152, 2012

Key words: **bone, metastasis, relapse, treatment, vaginal cancer**

Riassunto

Presentiamo il caso di una paziente di 71 anni con cancro squamoso primario della vagina (G3, FIGO I). È stata trattata con la resezione del tumore seguita da radioterapia. Quattro mesi dopo il trattamento completo le fu diagnosticata la recidiva del cancro nella colonna lombare. Non c'erano metastasi nella zona addominale o pelvi né in altri organi. La paziente ha ricevuto radioterapia palliativa per la colonna lombare. Nonostante il trattamento, la paziente è morta dopo 17 mesi dalla diagnosi del tumore primario a causa della progressione della malattia. Questo caso dimostra che nelle pazienti trattate per cancro vaginale primario, che presentano mal di schiena cronico bisogna essere particolarmente attenti, anche quando il cancro vaginale è stato diagnosticato in una fase clinica precoce. Eur. J. Oncol., 17 (3), 149-152, 2012

Parole chiave: **ossa, metastasi, recidiva, trattamento, cancro vaginale**

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Introduction

Metastatic bone tumors in women mostly originate from breast cancer, followed by lung, kidney and thyroid cancer (1). Bone metastases from gynecologic cancers are infrequent and are generally described in cases with a poorly differentiated (G3) high stage primary tumor, with local failure and indications of recurrent disease (2, 3). The lesions tend to be focal and osteolytic, rarely osteoblastic, and the mode of spread to bones in gynecologic epithelial cancers may be varied (1).

Metastases from gynecologic cancers have been described in all bones, with vertebral bodies being the most common site, followed by the ribs, clavicle, skull and femur (2-4). A review of the Medline base using the search terms cancer of the vagina and bone metastases showed only three such cases, but none when the cancer metastasized to the lumbar vertebrae (5-7). To the best of our knowledge, this is the first reported case of localized metastasis to lumbar vertebrae, arising from an early-stage cancer of the vagina.

Case report

A 71-year-old patient presented in March 2002 to the Gynecological Cancer Outpatient Clinic at the Regional Cancer Center in Lodz with a 3-month history of vaginal bleeding. The patient, gravida 2, para 2, had ceased menstruation at the age of 53 years. She did not have decreased body weight and presented no other suspected general symptoms. The patient had no history of prior abdominopelvic surgery.

On physical examination, the patient was found to be in good general condition, but gynecological examination showed a solitary tumor 6 mm in size, situated in the lower one-third of the vagina on its posterior wall. A biopsy confirmed poorly differentiated (G3) squamous-cell carcinoma of the vagina (Figure 1). Clinical examination and cervical cytology showed no signs of epithelial pathology. Transvaginal ultrasound showed no signs of uterine and adnexal pathology. Chest X-ray and abdominopelvic CT revealed no pathological findings. Her disease was classified as clinical stage I according to FIGO 2009 staging and pT1 N0 M0 according to the TNM classification.

The patient was treated with local tumor resection with negative surgical margins. Because of the poor histological differentiation of the tumor (G3), external and intracavitary irradiation was administered as adjuvant until July 2002. The external pelvic irradiation was delivered to the pelvis by Co60

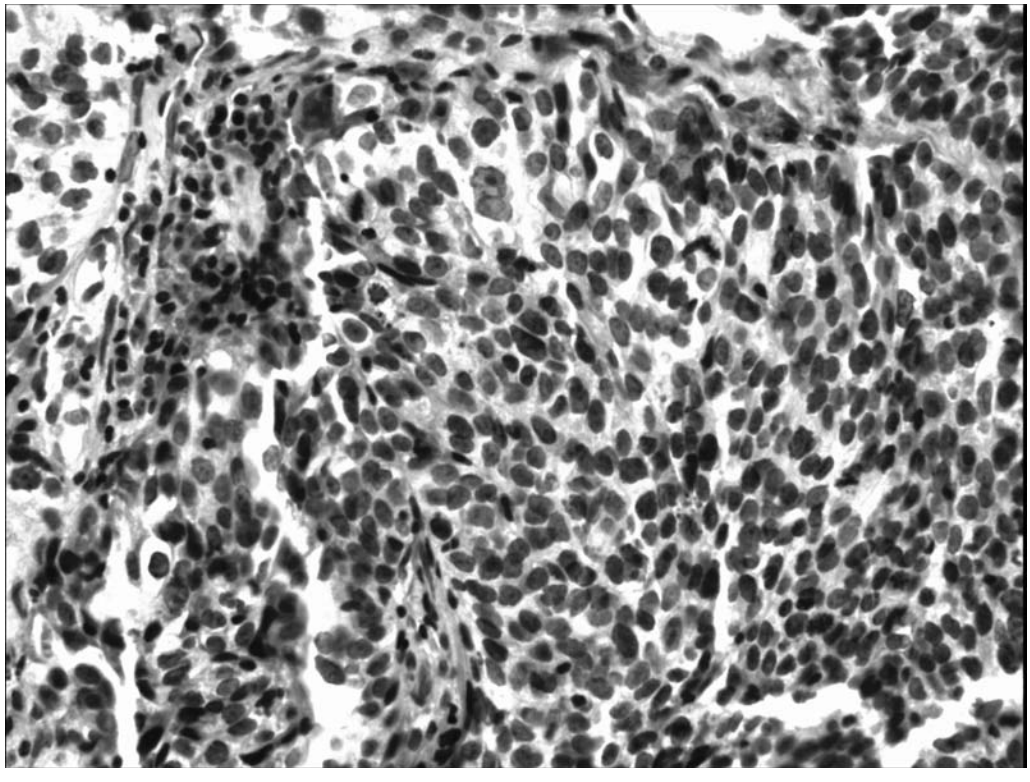

Fig. 1. Poorly differentiated (G3) squamous-cell carcinoma of the vagina – (HE, 200X magnification).

through four fields, and a total dose of 40 Gy was given in 20 fractions. The intracavitary treatment was performed during the external irradiation using a high-dose-rate Co 60 unit, with 18 Gy being delivered to the vaginal surface in 3 fractions.

The patient was disease-free and asymptomatic during the first 4 months after treatment. However in October 2002, pain of the lumbar area developed. X-ray showed metastasis to the lumbar spine with pathological fractures in vertebrae L2-L5. Bone scintigraphy demonstrated hot spots in the lumbar vertebrae (Figure 2) without any other skeletal lesions. No synchronous extra-osseous metastases were found. The patient underwent irradiation by Co 60 up to a total dose of 8 Gy, and received opioid treatment for pain (fenthanyl transdermally combined with morphine orally). The treatment was well tolerated and relief of symptoms was observed. She was discharged from the hospital. After four months, metastases in the liver were detected. The liver biopsy confirmed metastatic squamous-cell carcinoma. Unfortunately, the general condition of the patient quickly worsened making oncological treatment impossible, and only palliative treatment was administrated. The patient died in August 2003 due to progression of neoplastic disease.

Fig. 2. Whole body bone scintigraphy demonstrating hot spots in the lumbar spine.

Discussion

Our report presented in the previous section shows three main peculiar features: 1) a rare fatal outcome to primary cancer of the vagina diagnosed at clinical stage I, 2) the extremely rare occurrence of the relapse of cancer presented as osseous metastasis, 3) the bone metastasis detected as a single lesion with no other synchronous osseous or non-osseous metastases, which is an unusual feature in such cases.

Primary cancer of the vagina is rare, accounting for 2% of all gynecologic malignancies and 0.15% of all cancers in women (8). In the FIGO Annual Report, Beller et al. found only 324 cases worldwide in the years 1999-2001 (9). In keeping with the presented case, the disease is predominantly diagnosed in elderly women, and in 70-80% of cases occurs in patients above 60 years (8). The overall 5-year survival in patients is approximately 50%, but above 80% for clinical stage I (8), and the most common histology, as in the presented case, is squamous-cell carcinoma (9).

The treatment choice in patients with vaginal cancer is based on the medical condition, extension of the disease, location of the tumor in the vagina as well as clinical stage of disease, and traditionally includes radiotherapy and/or surgery in selected cases (5, 8-10). In our case, the location within the vagina, the size of the tumor and the lack of metastatic lesions made the complete resection of the primary cancer with adjuvant radiotherapy as the treatment of choice. Nowadays, it is postulated that patients with poorer prognostic factors as increasing stage, large tumors greater than 4 cm, involvement of regional lymph nodes and older age should be considered for treatment with additional chemotherapy (9, 10). In the described case, weekly cisplatin was not administered with the adjuvant radiation, but in 2002 it was not a preferred treat-

ment modality in our center. In the presented case, a good prognosis for the patient was given and chemotherapy was not administered. Unfortunately, an early osseous relapse developed 4 months after treatment of primary cancer. There is a lack of data to compare the time from the diagnosis of the primary vaginal cancer until bone metastasis, but in our case it was 7 months.

In advanced vaginal cancer, distant metastases are usually diagnosed in the lungs and in the liver (8); osseous metastases are extremely rare (5-7). In 1991, Corey et al. reported a case of metastasis from vaginal cancer to the temporal bone (6). Another location of osseous metastasis was recently described by Tjalma and Somville: the authors reported a case of a 74-year-old patient with a metastasis to the fibula as the presenting feature of vaginal cancer, who was surgically treated and, afterward, with radiotherapy of the metastatic lesion (5). A case of bone metastasis from vaginal cancer was reported also by Dutka et al. among 62 patients with bone metastases operatively treated due to pathological fractures (6). No other descriptions of osseous locations of relapsed squamous-cell carcinoma of the vagina may be found in the Medline database.

Despite the rarity of bone metastases from cancer of the vagina, there is a need to have a high index of suspicion for metastasis in patients with a history of cancer who present with osseous pain not responding to conservative treatment. The initial diagnosis can be challenging, as the symptoms are often attributed to other more common benign conditions such as soft tissue inflammation, trauma, arthritis, and osteomyelitis. Appropriate imaging may include plain X-ray photography and radionuclide bone scans (2-4). Therefore, in patients with suspected lesions, and who demonstrate evidence of bone destruction, a biopsy should be performed (2). CT scanning and nuclear magnetic resonance may help in dubious cases (3, 4).

The treatment strategy in patients with metastatic lesions in bones still remains a topic of controversy because of the few descriptions available in the literature and the different bone sites involved. For these reasons, a common suitable treatment regimen cannot be established and treatment should be tailored to each patient. Treatment by irradiation, with or without surgery and chemotherapy, is reported as effective in most cases and may be curative (3, 4). The main goal of treatment should be to eliminate or palliate pain and prolong survival (2). Generally, in patients with bone metastases from gynecologic cancers, despite treatment, the average duration of survival after relapse ranged between 7-12 months (1, 2, 4). In the presented case it was 10 months.

We conclude that patients presenting a history of primary cancer of the vagina with chronic pain in the vertebral column should be carefully evaluated, even if the cancer is diagnosed at an early clinical stage.

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