Solitary Metastasis in the Tibia as a Feature of Primary Squamous Vaginal Carcinoma: A Case Report

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Abstract. Aim: Primary vaginal cancer is rare, comprising about 3% of all gynaecological malignancies. Solitary bone metastases to appendicular skeleton are extremely rare and there are only few published reports. We report the case of isolated metastasis in the tibia as a feature of primary squamous vaginal carcinoma. We present this case because of its rarity and for documentation and discussion purposes. Case report: We present a case of a 44-year old woman diagnosed with squamous vaginal cancer in June 2015. Computed tomography of the thorax, abdomen and pelvis revealed no distant metastases. She was treated with interstitial intracavitary brachytherapy and concomitant chemoradiotherapy. In March 2016 she was admitted to our hospital because of the swelling and the pain in the lower right leg. There was no history of trauma and plain radiographs were normal. Two months later, after further progression of pain, plain radiographs showed intramedullary permeative bone lesion with cortical disruption of tibial diaphysis and local soft tissue swelling. MRI and core needle biopsy confirmed vaginal cancer metastasis containing tumor cells identical to the primary tumor. The patient was treated with radiotherapy and chemotherapy. Despite the treatment there was further progression of the disease with multiple bone metastases and eventually lung and brain metastases. Conclusion: This case report is another reminder that although extremely rare, solitary metastatic bone lesions of vaginal cancer are possible. Since symptoms mimic various benign conditions, it is important to consider bone metastasis as possible diagnosis in patients with progressive bone pain not responding to analgesic treatment.

Keywords: Diagnostic imaging; Neoplasm Metastasis; Vaginal Neoplasms

Despite metastasis to bone is common in solid tumors, the prevalence of solitary bone metastasis in vaginal carcinoma is unknown but is probably extremely rare due to few documented cases.

**INTRODUCTION**

We report the case of isolated metastasis in the tibia as a feature of primary squamous vaginal carcinoma. This is one of the few documented cases of metastasis to appendicular skeleton. We present this case because of its rarity and for documentation and discussion purposes.

**CASE REPORT**

A 44-year-old woman was diagnosed with vaginal cancer in June 2015. On gynecological examination, an exophytic 50 mm lesion was seen on the posterior vaginal wall, which bled easily on contact. There were no palpable inguinal lymph nodes. Biopsy taken from the lesion was reported as poorly differentiated squamous carcinoma. Magnetic resonance imaging (MRI) revealed an extensive neoplasm of posterior and lateral vaginal walls infiltrating the pelvic diaphragm with unilaterally enlarged lymph nodes of the obturator group. There was no evidence of the involvement of the cervix. No distant metastases were seen on subsequent staging computed tomography (CT) of the thorax, abdomen, and pelvis. The patient underwent interstitial intracavitary brachytherapy and concomitant chemoradiotherapy. The follow-up MRI showed small residual tumor and inapparent obturator lymph nodes. We have to inform about patient being exposed to diethylstilbestrol. In March 2016 the patient was admitted to our clinic complaining of progressive pain and swelling of her lower leg. No history of trauma was reported. Plain radiographs of the lower leg were taken, showing no pathomorphological change (Figure 1). Two months later, after further progression of pain plain radiography showed intramedullary permeative bone lesion with cortical disruption of tibial diaphysis and surrounding soft tissue thickening (Figure 2). MRI showed an extensive soft tissue mass with large extraosseous component with marked signal enhancement after administration of intravenous Gadolinium based contrast. The ADC values were low indicating high cellularity of the lesion (Figure 3). Additionally, PET-CT scan was obtained, showing no other distant metastasis. Core needle biopsy of the lesion described above was performed and pathohistological report confirmed metastatic lesion containing tumor cells identical to the primary tumor. The patient underwent radiation therapy and chemotherapy with 5-Fluorouracile after multidisciplinary team (oncologist, orthopedic surgeon, radiologist) consensus. The follow up MR in August 2016 revealed the same size of the metastatic bone lesion in the tibia but with decreased cellularity indicating favorable response to treatment. On the same day MR of the pelvis revealed bone lesions in both ischial tuberosities highly suspicious of metastases. Bone scintigraphy performed in September 2016 confirmed the suspected metastatic lesions and revealed a new lesion in the right calcaneus. In October 2016 the patient underwent surgery for the clinically evident metastasis in the right labia majora which was subsequently confirmed on pathohistological analysis. She was treated with chemotherapy (paclitaxel and carboplatin). Follow up PET/CT in November 2016 revealed the progression of known bone metastases and several new osseous lesions subsequently treated with palliative radiotherapy and Disodium pamidronate. During 2017 she received 6 cycles of chemotherapy with 2 separate cycles of palliative radiotherapy over ischial tuberosity and right femur. Despite the treatment there was further progression of the disease confirmed on follow up PET/CT studies performed in January and October 2018 with multiple bone metastases and eventually lung and brain metastases. The patient died little over three years after the initial diagnosis.
DISCUSSION

Vaginal cancer is a rare entity comprising about 3% of all gynecologic cancers. The most common histopathological type is squamous cell carcinoma mainly affecting postmenopausal women with peak incidence at age older than 70 years being followed by adenocarcinoma. The adenocarcinoma commonly affects younger women (median age, 19 years) and is more likely to metastasize to the lungs and lymph nodes. Moreover, the rising incidence of invasive vaginal cancer in younger women is in strong association with human papillomavirus (HPV) infection (93%)\(^1\). A subtype, clear cell adenocarcinoma, is associated with in utero exposure to diethylstilbestrol and is found in 2% of exposed females\(^1\).
As other gynecologic malignancies, vaginal cancer is classified according to the International Federation of Gynecology and Obstetrics (FIGO). MRI, which is not included in FIGO classification, has been shown to accurately contribute to diagnosis, local staging and dissemination of disease in vaginal cancer\(^7\). Other imaging modalities, such as FDG-PET and CT are useful for staging, follow up and treatment planning\(^2\).

CONCLUSION

Despite metastasis to bone is common in solid tumors, the prevalence of solitary bone metastasis in vaginal carcinoma is unknown but is probably extremely rare due to few documented cases. Even more rare is further progression to bones without another organ metastasis. Since symptoms mimic various benign conditions, it is important to consider bone metastasis as possible diagnosis in a patient with progressive bone pain not responding to analgesic treatment.

Traditional patients with bone metastasis have advanced stage disease, and the utility of surgical intervention is unclear. Some advocate minimal intervention and aggressive pain control, whilst others support more aggressive surgical intervention due to the unresponsiveness of these metastatic lesions to chemotherapy, radiotherapy and other noninvasive measures\(^6\). Due to the rarity of bone metastasis from gynecologic malignancies, the prognosis of these patients after surgical treatment has not been well established\(^6\).

This case report is yet another alert that although extremely rare, isolated metastatic bone lesions of vaginal cancer are possible and in the appropriate clinical setting should not be missed.

Conflicts of Interest: Authors declare no conflicts of interest.

REFERENCES


