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Commentary

Recognizing Rare Metastatic Presentations of Vulvar Cancer to Bone and Brain: A Short Review

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Abstract

Distant metastasis of vulvar cancer beyond the pelvis is rare. Overall prognosis in distant metastasis is poor and data on primary treatment in these cases are limited due the uncommon nature of the advanced disease. This article provides a brief overview of current literature on distant metastasis in vulvar cancer focusing specifically on metastasis to bone and brain or other neural tissue. A short commentary will also be provided on a recently published case report by the authors of this paper.

Keywords: Bone metastasis; Distant metastasis; Vulvar cancer

Review

Vulvar cancer accounts for a small percentage of new cancer cases in the United States making it an uncommon gynecologic malignancy. Some have suggested that anywhere between 5 and 8% of all gynecologic malignancies are vulvar in origin [1]. Prognosis of vulvar cancer depends on the surgical pathological stage, histologic differentiation and lymph node involvement. For the purpose of this paper distant metastasis is defined as anything beyond the pelvis or pelvic lymph nodes including groin nodes. Distant metastasis in vulvar cancer has a poor prognosis and rare based on limited number of studies and reports. In a multicenter study of 502 patients, distant metastatic recurrences revealed a five year survival rate of 15% with lymph node metastasis being the primary predictor for recurrence [2]. If metastasis does occur beyond the pelvis, the most frequent sites are liver, lung, brain and skin with a few case reports demonstrating involvement of kidneys [3,4].

One review looked at 391 patients with squamous cell cancer of the vulva; only 5% of patients were found with distant metastasis. Of those with distant metastasis, 9 had lung involvement followed by 7 with liver, 5 with bone and 4 with skin metastasis [5]. Another retrospective study identified 1686 patients with epithelial gynecological cancers and of those, patients, only 18 patients had bony metastasis. Overall, metastasis to bone occurring in only about 1% of gynecologic malignancies and most common bone site was the vertebrae. Furthermore, only one case in the entire study demonstrated bony metastasis from a primary vulvar malignancy [6].

Another distant site of metastasis in vulvar cancer that has been infrequently reported, is the brain. Brain metastasis in gynecologic malignancy is uncommon with highest incidence occurring in choriocarcinomas [7]. There have only been few case reports of brain metastasis specifically in vulvar cancer [7,8]. Prevalence is unknown and prognosis is quite poor. Data on systemic treatments is limited for patients with distant metastatic vulvar cancer due to rarity of disease. Primary treatment for distant vulvar metastasis can include radiation therapy, systemic chemotherapy and palliative treatment. There is no standard of care due to the low incidence and lack of reported vulvar cancer with distant metastasis. A platinum-based combination chemotherapy is most commonly administered. According to the National Comprehensive Cancer Network, current guidelines and recommendations are supported by evidence extrapolated from treatments related to cervical, anal and other squamous cell cancer treatments, however the evidence supporting these recommendations are not specific to vulvar cancer. Larger randomized controlled trials are lacking and needed in order to give best evidence-based guidelines for recurrent vulvar cancer with distant metastasis.

Commentary

A recently published case report titled “Rare phalanges soft tissue and bony metastasis in vulvar squamous cell carcinoma: Case report” showed distant metastasis to both brain and bone [9]. The case report was about a 52-year-old with an initial diagnosis of vulvar squamous cell carcinoma treated with radical surgery, chemotherapy and radiation according to National Cancer Center Network guidelines. Despite a 14-month window of remission, she developed aggressive recurrence with atypical metastatic sites of the clavicle, phalanges, and brain. She presented with bleeding from her finger and underwent amputation. Pathology revealed metastasis consistent with her primary vulvar cancer. She also required craniectomy, stereotactic radiation, and systemic chemotherapy, and later had a pathologic fracture of the left clavicle due to metastatic disease. She subsequently was transitioned to palliative care.

This was the first case describing soft tissue finger metastasis from vulvar cancer requiring amputation. To our knowledge, this case report was also the first to show both bony and brain metastasis from a primary vulvar malignancy. The report provides a concise clinical

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presentation and treatment timeline of an uncommon case of distant metastasis in vulvar cancer along with therapeutic challenges. Additional studies are needed to determine the best treatment options for metastatic disease before guidelines are established.

Conclusion

The case impresses the need for surveillance and comprehensive monitoring and patients with vulvar cancer during remission. While rare, uncommon metastatic sites should be in the differential for patients presenting with abnormal complaints with history of vulvar cancer. Thorough physical examinations and soft tissue biopsies should be emphasized and considered for any suspicious lesions, regardless of how uncommon the metastatic site may be. This case report adds uncommon presentations of distant metastasis of vulvar cancer to the literature, thereby aiming to improve patient outcomes and quality of life.

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