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Case Report

Bone metastasis from malignant phyllodes of the breast

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ABSTRACT

Cystosarcoma phyllodes was described first in 1838 and originally was considered to be a benign tumor. It was not until 1931 that metastasis from a cystosarcoma phyllodes was reported. The incidence of cystosarcoma phyllodes is estimated to be 0.3% to 0.9% of all breast tumors. Sites most commonly affected by metastases are the lungs and bones. We present a case report 29-year old female patient presented with a voluminous breast mass at both of them which was completely resected. The right side presented of malignant phyllodes and the left side is borderline phyllodes. Six months later, both of her legs became paralyzed and accompanied by swelling over her right upper arm. Biopsy was performed, and the diagnosis was metastatic malignant phyllodes tumor. Histologic review of the breast tumor revealed stromal overgrowth.

Keywords: Bone metastasis, Breast tumor, Malignant phyllodes, Stromal overgrowth

INTRODUCTION

Cystosarcoma phyllodes was described first in 1838 and originally was considered to be a benign tumor. It was not until 1931 that metastasis from a cystosarcoma phyllodes was reported. The incidence of cystosarcoma phyllodes is estimated to be 0.3% to 0.9% of all breast tumors.[1] Phyllodes tumors (PT), previously called cystosarcoma phyllodes, are fibroepithelial tumors of the breast. PT are uniquely found in breast tissue and histologically have both an epithelial component as well as metaplastic mesenchymal (stromal) component. Clonal analyses have revealed that some PT develop from fibroadenomas. At least 12.5% of patients with PT have a history of fibroadenomas and over 20% of patients have a concurrent diagnosis of a benign fibroadenoma. The pathological appearance of stromal overgrowth, stromal cellularity, degree of nuclear atypia, number of mitotic cells, and the nature of the tumor margin (circumscribed, pushing, or infiltrative) determines whether a tumor is a benign fibroadenoma, a benign PT, a borderline PT, or a malignant PT. The stroma from malignant PT are distinguished by marked cellularity with nuclear atypia, nuclear pleomorphism, increased mitotic activity, and stromal cell overgrowth. Heterogeneous stromal components are seen in PT and is commonly associated with malignant PT. Malignant PT has a 5- and 10-year overall survival of 84% and 77% after surgical extirpation.[2] Studies show that a malignant course may be seen in 15-30% of the cases, with metastatic spread occurring in about 5%.[3] Most distant metastases develop without evidence of local recurrence. The commonest sites for distant metastases are the lung, bone, and abdominal viscera. These often occur in the absence of lymph node metastases and histologically contain only the stromal element.[4]

CASE REPORT

There were 29-year old female patient complains of back pain from 1,5 mo prior to admission and become heavy in the last 12 days. Both of her legs became paralyzed. She also presented with swelling on the right upper arm over 2 months (Figure 1), and it became more severe if it moves. Patient had a lump at the right breast 4 years prior, and a similar symptom and the left side 3 years

prior. The patient had undergone Modified Radical mastectomy for malignant phyllodes tumor 6 month prior and left side simple mastectomy for borderline phyllodes tumor 3 months prior (Figure 1).

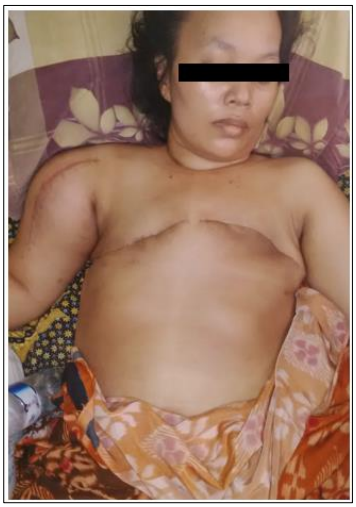

Figure 1: Clinical picture of patient.

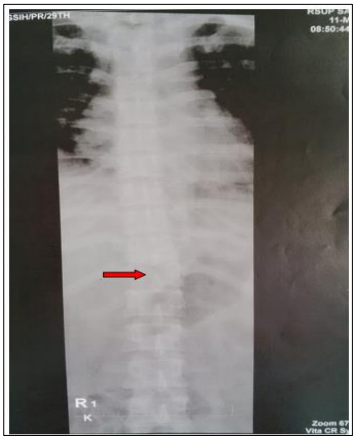

Figure 2: AP radiograph of thoracolumbar spine demonstrating lytic destruction of Th XI.

Histologic review of the breast tumor revealed malignant phylloides tumor at the right side and borderline phylloides tumor at the left side. We got a lytic lesion at the vertebrae thoracal IX (Figure 2) and 1/3 middle of right humerus from x-ray (Figure 3).

We suspected with a pathological fracture. Patient had undergone a decompression stabilization, fusion and biopsy for the Thoracal vertebrae (Figure 5), and the histological review with a stromal overgrowth which is a metastatic process from malignant phylloides tumor (Figure 4). Patient also already had undergone an ORIF PS (Figure 6) and biopsy for the right humerus. The histological review a same process above. The patient had a karnofsky score 50, in that case we do not give any chemotherapy and radiotherapy.

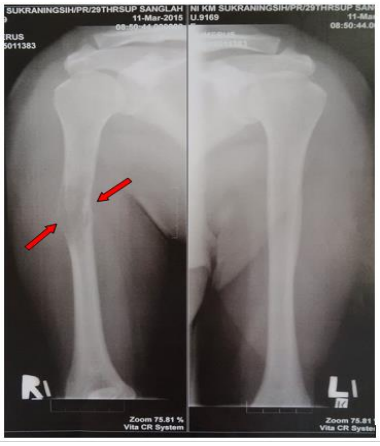

Figure 3: AP radiograph of right and left Humerus demonstrating lytic destruction of 1/3 middle of right humerus.

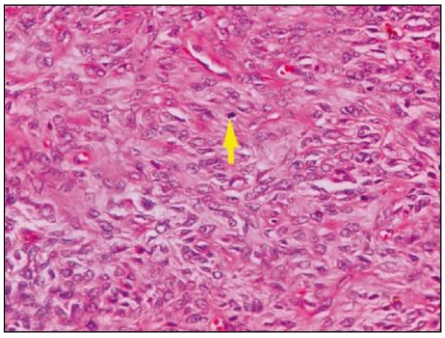

Figure 4: Malignant phyllodes tumor showing marked stromal overgrowth, nuclear atypia, and mitosis (arrow) (H and E) ×400.

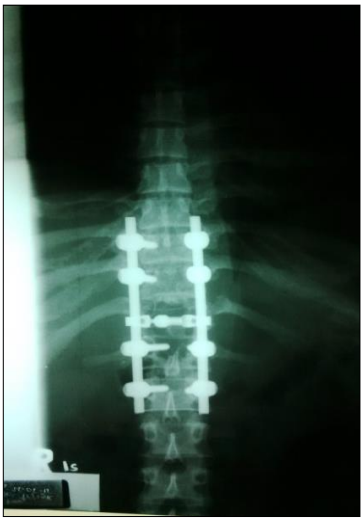

Figure 5: AP radiograph of thoracolumbal spine post decompression and posterior spine fusion.

DISCUSSION

The characteristics of a unique fibroepithelial neoplasm were first noted by the renowned surgeons Sir Astley

Cooper and Sir James Paget, who described their experiences with "cellular hydatids" or "glandular proliferous cysts". Nevertheless, it was not until 1838 that Muller systematically collected several specimens of an unusual breast tumor characterized by large bulk, rapid growth, and peculiar gross and microscopic features. He considered this entity a genuine neoplasm and designated it cystosarcoma phyllodes, not to imply malignancy, but to describe the fleshy appearance of the tumor. Since then, its clinical behavior and malignant capacity have been the subject of several studies.[5]

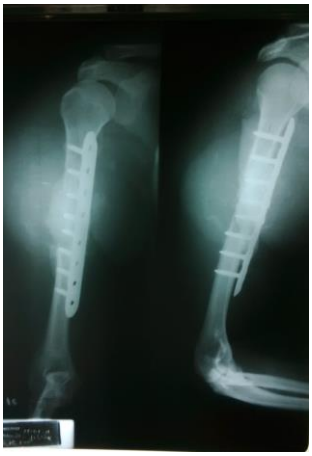

Figure 6: AP/Lat radiograph of right humerus post ORIF PS.

Initial attempts to identify the more aggressive sub type of PT based on histological grading yielded limited result. Recently, however, the histologic feature of stomal overgrowth has been note to correlate closely with potential for metastatic spread. In several series, the tumor of all patients who subsequently developed metastasis demonstrated this feature of stromal overgrowth.[6]

Malignant PT with differentiated sarcomatous components as a portion of the stromal tumor have been reported: osteosarcoma, liposarcoma, leiomyosarcoma, fibrosarcoma, chondrosarcoma, rhabdosarcoma and malignant fibrous histiocytoma. In a review of all reported cases of metastatic PT (67 cases) prior to 1972, Kessinger and colleagues noted that the average survival time after diagnosis of metastasis was 30 months. Metastatic lesions have been reported as early as at the initial diagnosis of the primary tumor and as late as 12 years after diagnosis. The longest survival time after diagnosis with metastatic disease was 14.5 years. The most common site of the initially diagnosed distant metastasis was the lungs. Other metastatic sites include the bones, liver, heart, and distant lymph nodes.[7]

The diagnosis of metastatic malignant phyllodes tumor was made in the basis of the malignant stromal material obtained on FNA. As reported by some authors, it is the malignant stromal component that metastasizes. Only rarely has the epithelial component been reported in metastasis.[8]

The FNA findings of the metastatic tumor consisted of a hypercellular aspiration of sarcomatous spindle cells arranged singly and in fascicles and exhibiting hyperchromatic and pleomorphism. The cell block preparation of the metastatic tumor had a cellular pattern similar to that of the primary breast tumor.[9]

Whereas both the stromal and epithelial cells were present in the breast tumor, only the stromal component was seen in the metastatic lesion.[10] Histopathology of the original breast tumor revealed many areas of stromal overgrowth, a feature which is strongly correlated with subsequent metastatic activity.[11]

Moreover, those patients with malignant phyllodes tumors have a 20-25% incidence of distant metastasis. A recent study of 48 women established that distant metastasis developed in 37% of the cases, 30% following local excision, 14% following wide local excision and 33% after mastectomy.[8]

Regardless of grade, the primary method of treatment is surgical. Options include local excision, wide local excision and mastectomy. Wide excision is recommended because of the strong tendency for local recurrence (up to 20%), even in benign PTs. In a study of 172 patients with PT, local excision was defined as removal of the mass with no more than a 0.5 cm tumor free margin while wide local excision was defined as removal of the mass with at least a 1.0 cm tumor free margin.[9] Of the 172 patients, 76% were benign, 7% were borderline and 17% were malignant. There was a tendency for less aggressive surgical treatment of lower grade tumors with mastectomy performed in 10% of benign cases, 67% of borderline cases and 86% of malignant cases.[12] Local recurrence occurred only in the benign cases treated with local or wide local excision with a rate of approximately 15%. When metastatic, lymph node involvement is rare with authors noting a rate of approximately 10%.[8]

Metastases from cystosarcoma phyllodes are rare, and treatment generally is ineffective.[13] The rarity of metastatic cystosarcoma phyllodes makes it difficult to do systematic trials.[10] The efficacy of systemic chemotherapy against metastatic MPT remains unknown. It is based on the principles of sarcoma rather than carcinoma treatment. There are relatively few case reports of single agent chemotherapy. Doxorubicin and Dacarbazine used as single agents have yielded no satisfactory response.[11]

Hormonal therapy is not effective in phyllodes tumor. This is despite the presence of positive hormone receptors. The hormone receptors are component of the epithelial aspect of the tumor whereas the stromal aspect of the tumor is responsible for the metastatic behavior of the tumor. The receptor from the stromal component

expresses estrogen receptor beta, not alpha as with adenocarcinoma. To date, there is no established role for adjuvant chemotherapy or hormonal therapy in phyllodes tumor.[14]

Increased length of survival, definitive operative stabilization should be considered in addition to adjuvant therapy in patients with cystosarcoma phylloides.[5]

Two side metastatic in one time make this is rare case. Despite of this is the first case that been reported.

Since these tumors are rare, and because of the lack of a consensus on the appropriate treatment modality for these neoplasms, such cases must be referred to highly experienced specific institutions when encountered.[15]

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