RESEARCH ARTICLE

CASE REPORT: METASTATIC PHYLLODE TUMOR

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Abstract

Rationale: Malignant phyllodes tumors are rare breast neoplasms that are associated with a 6.2% to 25% incidence rate of distant metastasis; the lung is the most common metastatic site. To date, there is no consensus regarding the treatment of metastatic malignant phyllodes breast tumors.

Patient concern: A 34-year-old woman was admitted into the gynecology department for a rapidly growing left breast tumor that was first noticed month prior.

Diagnosis: Core needle biopsy revealed a malignant phyllodes tumor. A chest computed tomography/CT showed metastatic lymph nodes that appeared to have spread to the right axilla. She was subsequently followed by course of radiotherapy, she consulted again 3 months later for a productive cough. X-ray thorax in comparison with that made preoperatively: presence of the left peri-hilar nodules which were not present on the first X-ray. Interventions: A left mastectomy with axillary lymph node dissection was conducted and a thoracoabdominal flap and a split thickness skin graft were performed for the skin defect. And radiotherapie adjuvant.

Lessons: As standard treatment guidelines for metastatic malignant phyllodes tumors are lacking, we opted for the aforementioned aggressive treatments that resulted in complete remission of the lung metastasis. Therefore, aggressive treatment, whenever possible, is warranted.

Introduction:

Breast cancer accounts for the most frequently diagnosed malignancy and the Second-highest number of cancer-related deaths in women. Although the most Common types of breast cancer are epithelial tumors of the ducts of lobular units, other types exist which affect mortality significantly. Another subtype category is the family of fibroepithelial tumors, which include fibroadenomas most commonly and phyllodes tumors. Phyllodes tumors account for 0.3 to 1% of all breast tumors and 2.5% of all fibroepithelial tumors of the breast [1].

Though phyllodes tumors are similar to fibroadenomas in pathology, they have a double-layered epithelial component surrounded by an increased stromal hypercellular component, forming leaf-like processes [2]. The median age of onset for phylloide tumors is 45 years [3]. In 1981 the World Health Organization adopted the term phyllodes tumor and sub-classified them histologically as benign, borderline, or malignant according to the features such as tumors margins, stromal overgrowth, tumor necrosis, cellular atypia, and number of mitosis per high power field as described by Rosen in Rosen’s Breast Pathology, Lippincott. Reports suggest that approximately 85–90% of
Phyllodes tumors are benign and 10–15% were malignant [4]. Only 10-26% of malignant phyllodes tumors are found with metastasis [5–6]. While difficult to identify, they are generally characterized by rapidly growing huge masses that may present with delayed metastases, mainly to the lung [7]. Surgery with sufficient resection margins is the standard treatment for phyllodes tumors, and adjuvant radiotherapy is recommended to reduce local recurrence.

**Case Presentation**

We report the case of a 34-year-old woman, single, nulligeste, who consults for a rapidly growing left breast tumor that was first noticed 2 months prior. On physical examination, a massive tumor occupying the entire left breast was observed. The skin had been stretched thin, with areas of necrosis as well as engorgement with a stretched superficial vein (Fig. 1). There were no abnormal findings in the patient or family records. Breast ultrasound showed a massive tumor with cystic components occupying the entirety of the left breast. Core needle biopsy revealed a malignant phyllodes tumor. Computerized tomography (CT) of chest, abdomen and pelvis showed no lung or liver or others metastasis. A left mastectomy with axillary lymph node dissection was conducted and a thoracoabdominal flap and a split thickness skin graft were performed for the skin defect (Fig 2). She was subsequently followed by a course of radiotherapy, she consulted again 3 months later for a productive cough. X-ray thorax in comparison with that made preoperatively: presence of the left peri-hilar nodules which were not present on the first X-ray, a CT (thoracic) returning in favor of a suspect lower right lobe pulmonary nodule measuring 12mm, she benefited from a lower right lobectomy. Anapath: in favor of a secondary localization of phyllodes sarcoma.

**Figure 1:** A giant phyllodes tumor of LEFT breast in a 34-year-old woman.
Figure 2: Operative findings. Left mastectomy with axillary lymph node Dissection; thoracoabdominal flap and a split thickness skin graft.

Discussion:
Phyllodes tumors occur in two types of breast tissues: stromal (connective) and glandular (lobule and duct). The stroma contains the neoplastic component and has the potential to metastasize. The standard treatment for phyllodes tumors is wide excision with sufficient surgical margins. Surgical margins of >1cm have been associated with lower local recurrence rates in borderline and malignant phyllodes tumors. However, securing a sufficient margin is difficult in most cases because of huge tumor sizes that can occupy the entire breast, and complete mastectomy is often required. However, skin that is stretched thin, exhibits ulcers, and/or has necrosis may also require extensive removal during surgery; this can lead to difficulties in primary suturing owing to extensive skin loss, necessitating a compensatory flap. In our case, a thoracoabdominal flap and split thickness skin graft were used after the mastectomy, and a surgical margin of ≥1cm was secured in all directions except the posterior margin. Hematogenous spread is the most common metastatic route, and the lungs is the most common site. Axillary lymph node involvement is rare, and routine axillary surgery is not recommended. However, in our patient, suspicious lymph node involvement accompanied by CTs, axillary dissection: 19 non-metastatic lymph nodes (19N- / 19N)

Though adjuvant radiotherapy remains controversial for PTs according to National Comprehensive Cancer Network (NCCN) guidelines for the management of PTs (version. 5, 2020), use of radiotherapy has increased recently due to high risk of recurrence. Adjuvant radiotherapy is recommended in borderline and malignant PTs with tumor-free margin < 1 cm. For high-risk malignant tumors (higher grade, size > 5 cm), adjuvant radiotherapy is considered after R0 resection and recommended after R1 resection.

The efficacy of palliative chemotherapy for metastatic malignant phyllodes tumor is unknown. Ifosfamide is considered the most active agent for metastatic malignant phyllodes tumors; doxorubicin and dacarbazine have been reported to be effective when administrated with cisplatin or ifosfamide. In our patient, lung metastasis are found after surgery; the decision to perform a lower right lobectomy. As standard treatment guidelines for metastatic malignant phyllodes tumors are lacking, we opted for the aforementioned aggressive treatments that resulted in complete remission of the lung metastasis. Therefore, aggressive treatment, whenever possible, is warranted.

The reliability of prediction of clinical outcomes based on morphological features (grade), even with clinical and radiological correlation, is poor. PTs are often misdiagnosed or mismanaged with dominant themes of under-diagnosis and under-treatment.
In conclusion:-
The phyllodes tumor is a rare mixed mesenchymal and epithelial primary breast neoplasm, which affects a younger cohort of patients than the more common primary epithelial derived breast adenocarcinoma. Depending on histopathological characteristics, the tumor behavior may range from benign with a similar surgical response compared to that of the fibroadenoma, to local aggressiveness to distant metastasis with associated poor prognosis. A key to the successful management of this tumor may be early detection and resection prior to the development of distant metastasis. Given the low incidence of this tumor and the current lack of specific clinical and imaging characteristics to make this diagnosis, more pooling of data may be beneficial to identify specific findings that increase pre-test probability prior to making a decision whether to observe the mass or proceed to surgery. Adjuvant chemotherapy is suggested for selected patients with large, high risk, or recurrent tumors only after thorough evaluation. However, no standardized therapy for treating these tumors exists to date.

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