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Case Report

Multidisciplinary Treatment for Recurrence of a Borderline Phyllodes Tumor: A Case Report

Hironaka-Mitsuhashi A*, Tabata R, Harada T, Nagata A

Fukuchiyama City Hospital, 231 Atunaka-cho, Fukuchiyama, Kyoto 620-8505, Japan.

***Corresponding author:** Hironaka-Mitsuhashi A, Fukuchiyama City Hospital, 231 Atunaka-cho, Fukuchiyama, Kyoto 620-8505, Japan.

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Abstract

Background: Phyllodes tumors (PTs) are rare neoplasms that are classified as benign, borderline, or malignant. Although borderline PTs are generally less likely to recur or metastasize than are malignant PTs, they often undergo malignant transformation during relapse. Resection is recommended for local recurrence, whereas current treatments for metastasis have not been established. The prognosis of metastatic and recurrent PTs is poor. Clarification of the postoperative monitoring methods for PTs, especially borderline PTs, is important, so that timely treatment can be initiated if needed. Herein, we present a rare case of a borderline PT in which skin and lymph recurrence was suspected early after radical surgery. **Case Presentation:** A 44-year-old woman with a borderline PT in her right breast was referred to our hospital. A mastectomy with axillary lymph node sampling was performed. Suggestive of recurrence,18F-fluorodeoxyglucose (FDG) positron emission tomography-computed tomography showed FDG accumulation in the skin and a lymph node shortly after surgery. Multidisciplinary treatment consisting of chemotherapy (with doxorubicin) and radiation therapy (like that for sarcoma) was administered, and the patient is alive >2 years thereafter. **Conclusions:** Our case suggests that early detection and medical intervention can control the recurrence of borderline PTs.

Keywords: PET/CT; Phyllodes Tumors; Recurrence; Lymph Node Metastasis;

Introduction

Background

Phyllodes tumors (PTs) are rare neoplasms that account for 1% of all breast neoplasms in women [1, 2]. They are classified as benign, borderline, or malignant, depending on histological features, such as margin involvement, cellularity, mitosis, and cellular atypia [1].

The standard treatment for primary PTs is complete excision. The National Comprehensive Cancer Network clinical practice guidelines recommend a surgical margin of =1 cm regardless of the type and size of the PT [3]. Advanced PTs require systematic treatments, which are generally based on those used for soft tissue sarcomas [3]. However, efficacy is low [4, 5], with a reported median overall survival time of 15.2 months (interquartile range: 7.6–39.6 months) [5].

Recurrence is associated with malignant and borderline PTs. Local recurrence is reported in 10–17% of benign PTs, 14–25% of borderline PTs, and 23–30% of malignant PTs; metastasis is reported in 0.13%, 1.62%, and 16.71%, respectively [6]. Although borderline PTs rarely metastasize, they often undergo malignant transformation upon recurrence [5, 6]. When metastasis does occur, the prognosis is poor [4-6]. Clarification of postoperative monitoring methods is urgently needed, so that timely treatment can be administered if needed, especially for borderline PTs.

Herein, we describe the multidisciplinary treatment of a borderline PT. Suggestive of skin and axillary lymph node recurrence, 18F-fluorodeoxyglucose (FDG) positron emission tomography (PET)-computed tomography (CT) early after radical surgery showed FDG accumulation at these sites.

Case Presentation

A 44-year old woman was admitted to a local clinic complaining of a painless fixed lump in her right breast. CT revealed that the caudal side of the right breast had nearly been replaced

with a tumor. A swollen right axillary lymph node was detected (Fig. 1a). She was referred to our hospital with a diagnosis of benign PT. The lump had grown rapidly in approximately 1 month. Her right nipple had lost its original shape owing to the tumor, and her skin of right breast had darkened (Fig. 1b). We decided to perform a mastectomy with axillary lymph node sampling as soon as possible. Skin grafts were used to close the wounds and ensure adequate surgical margins.

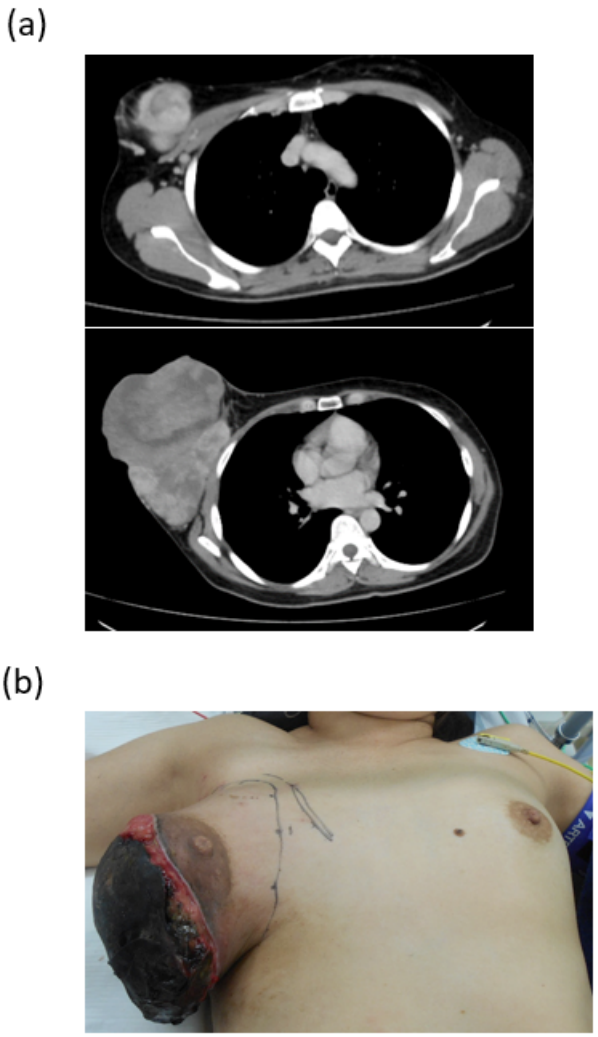

Figure 1: Clinical presentation. The tumor has rapidly grown. Preoperative computed tomography reveals replacement of the caudal side of the right breast by the tumor and an enlarged axially lymph node. (a, b) The patient's nipple appears enlarged and the skin darkened (b).

The tumor was well-defined with dimensions measuring 20 × 15 × 12 cm (Fig. 2a). Histologically, it exhibited a leaf-like pattern without invasion. A few atypical cells were also observed (Fig. 2b). Consistent with a borderline PT, Ki67 positivity was 15% in the epithelial lesions and 3% in the stromal lesions. The resection margins were free of tumor (Fig. 2a). Histological examination revealed no metastasis to the sampled lymph nodes.

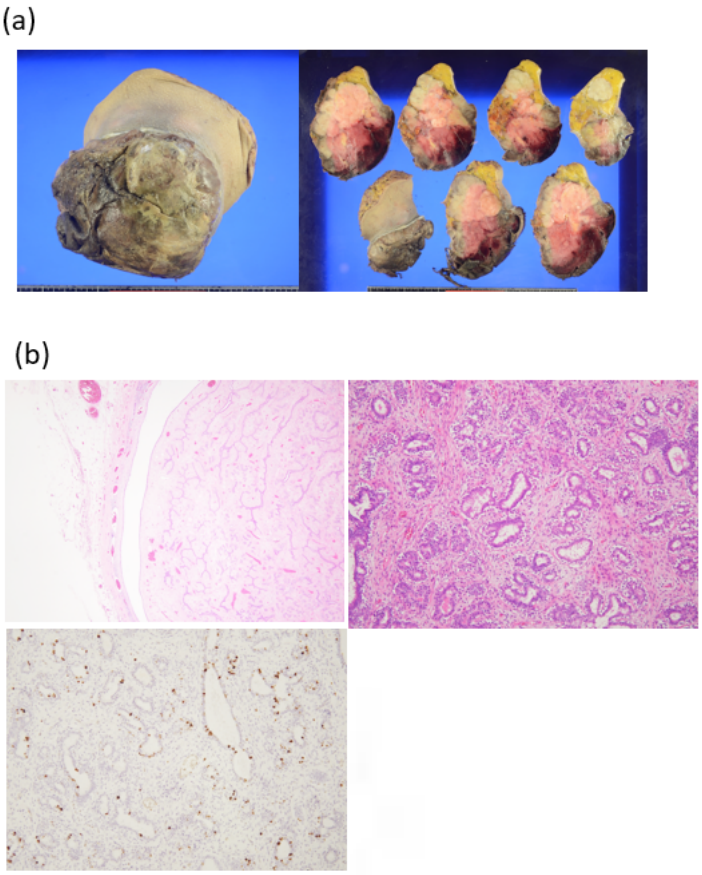

Figure 2: Histological examination. The margins are negative macroscopically (a). Microscopically, the tumor is consistent with that of borderline phyllodes tumors (b).

One month after the radical surgery, PET/CT revealed FDG accumulation in the skin and axillary lymph node (Fig. 3a). No redness or swelling was found on the skin. CT performed 2 weeks after PET/CT showed that the swollen lymph node had not shrunk (Fig. 3b). Recurrence was suspected, and radiotherapy was administered.

After radiotherapy, no new lesions appeared. However, the size of the lymph node had only slightly decreased (Fig. 3c); therefore, doxorubicin (60 mg/m2, 3-week cycle) was administered intravenously. CT revealed a further decrease in lymph node size after four cycles of doxorubicin; thereafter, the patient refused to continue chemotherapy.

The patient underwent a follow-up CT every 6 months. PET/CT 15 months after surgery revealed no accumulation of FDG in the previously indicated areas (Fig. 3d). The patient has survived for >2 years without any new recurrence after multidisciplinary treatment.

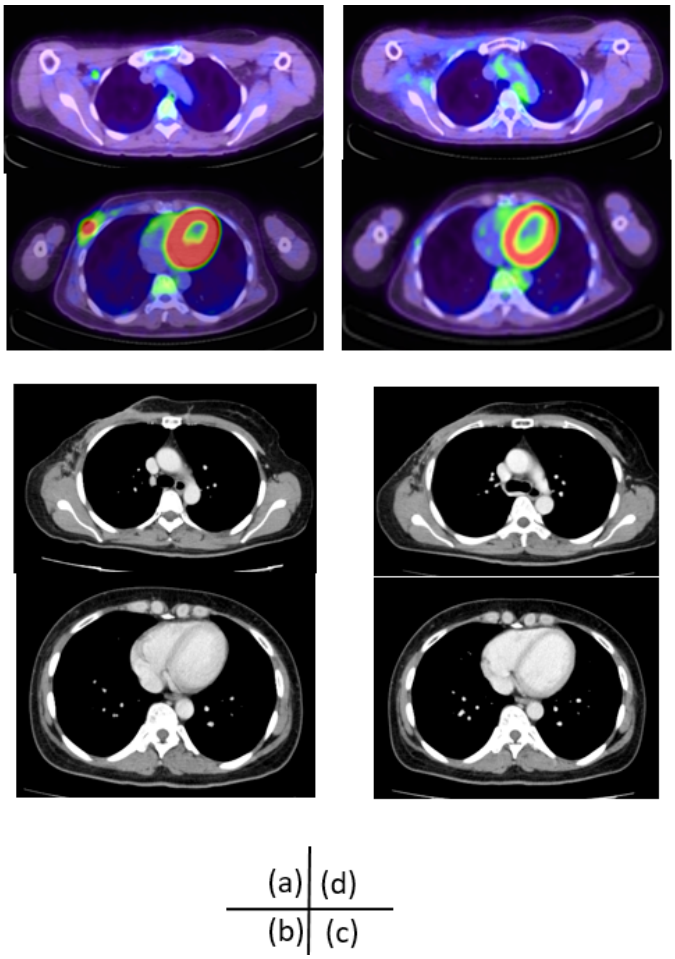

Figure 3: Change in lymph node size during treatment. 18F-fluorodeoxyglucose (FDG) accumulation is observed in the skin and axillary lymph node postoperatively (a). Computed tomography reveals that the size of lymph node has decreased after multidisciplinary treatment (b, c). FDG is no longer present 15 months after surgery (d).

Discussion

PET/CT is reportedly useful for detecting rare metastatic sites in PTs [7]. In our case, the question remains as to whether FDG accumulation actually indicated recurrence. Preoperative PET/CT was not performed because of the clinically rapid tumor growth; therefore, it was not possible to compare pre- and postoperative PET/CT images. Although FDG accumulation was observed in the skin and lymph nodes postoperatively, there were almost no gross or imaging findings at these surgical sites. In addition, no pathological examination was performed to determine whether the lymph node swelling was indeed due to metastasis.

The lymph nodes are rare sites if metastasis or recurrence are pointed out [8]. Therefore, axillary node dissection is not recommended for patients with PTs [2,3]. The time from diagnosis of a primary PT to the appearance of metastasis varies widely [5]. In the present case, preoperative CT revealed an enlarged axillary lymph node, and radical surgery with axillary lymph node sampling was performed. Pathological examination confirmed sufficient surgical margins and absence of axillary metastases. Considering the rate of tumor growth, lymph node recurrence may have occurred soon after surgery owing to inadequate lymph node procedures. The size of the lymph node decreased during multidisciplinary treatment. Thus, we believe that our case showed a rare type of recurrence.

Conclusions

Our patient was followed up without any treatment. Our case highlights the importance of early detection of recurrence and medical intervention.

Declarations

List of Abbreviations

CT: computed tomography

FDG: 18F-fluorodeoxyglucose

PET: positron emission tomography

PT: phyllodes tumor

Ethics Approval: This study was approved by the institutional review board of our institution (IRB No. 5-70).

Consent for Publication: Written informed consent was obtained from the patient for publication of this report.

Availability of Data and Materials: All data are available from the corresponding author upon reasonable request.

Competing Interests: The authors declare that they have no competing interests related to this case.

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