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Case Report

Vulvar metastasis from carcinoma breast unveiling distant metastasis: Exploring an unusual metastatic pattern

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Abstract A 76 year old woman with a previous history of infiltrating ductal carcinoma of right breast (diagnosed and treated 14 years back) presented to us with a non-healing ulcer on the left side of the vulva along with two satellite nodules close to the vulvar lesion. Biopsy showed an infiltrating ductal carcinoma of breast with a strong positivity for estrogen/progesterone receptors. Further, ^{18}F-FDG PET–CT (Fluoro-deoxy glucose positron emission tomography computed tomography) showed multiple bilateral lung metastases. She responded well to hormone therapy (Letrozole) with decrease in the size of primary vulvar lesion and disappearance of the satellite nodules. Repeating PET–CT at 6 months showed partial response of the lung lesions. The present case is unique in the way of metastatic presentation of breast cancer to vulva after a long gap of primary diagnosis (longest reported till date) and also in unveiling of further metastatic sites in otherwise asymptomatic case. Patients (particularly elderly) with this unusual and clinically isolated pattern of metastasis might remain misdiagnosed for a long period of time and this case report aims to increase the awareness of clinicians toward the same. Gynecological surveillance remains of paramount importance in the follow up of breast cancer.

Introduction

Breast cancer mostly metastasizes to regional lymph nodes, bones, lung, liver, brain etc. An unusual site of metastasis particularly to gynecological sites is rare and mostly involves ovaries or uterus [1]. Vulvar metastasis is a rare occurrence

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[2–4]. We report here an interesting and intriguing case of vulvar metastasis from a breast cancer unveiling further distant sites of metastasis occurring 14 years after the initial diagnosis.

Case report

A 76 year old female presented with a non-healing ulcer in the vulvar region for 1 year to our department in October 2014. She noticed a small elevated, reddish ulcer in the vulvar region, which was progressively increasing over the period of 1 year. She also complained of infrequent episodes of bleeding from the lesion. She visited local gynecologist for the same and had no relief with the course of antibiotics and medicines.

She is already a diagnosed case of carcinoma right breast (infiltrating ductal carcinoma; estrogen and progesterone receptor positive) in January 1999. She was treated with right modified radical mastectomy and pathologically was T2N0M0 as per AJCC staging [5] of cancer (tumor size: $3 \times 2 \times 1.5$ cm; 0/10 axillary lymph nodes positive). She also received 6 cycles of chemotherapy (Cyclophosphamide/Methotrexate/5-Fluoro uracil based chemotherapy) followed by 5 years of hormone therapy (tamoxifen) till July 2005. She was on regular follow up till 2010 and then defaulted for follow up owing to personal reasons.

On examination at this presentation, a $3\ \text{cm} \times 2\ \text{cm}$ ulcer was noted on the left side of mons pubis close to midline (Fig. 1a). The ulcer was non-tender and had a slough covered base with erythematous and indurated margins without any underlying fixity to deeper structures. Two satellite erythematous nodules (approximately 0.5 cm each) were also seen away from the primary lesion as evident in Fig. 1a. Left breast, right chest wall, bilateral axilla and supraclavicular examination revealed no signs of disease as were the systemic examination. Biopsy from the vulvar lesion was suggestive of infiltrating carcinoma of breast with strong positivity of estrogen and progesterone receptors (Fig. 2a–d). Whole body ^{18}F-Fluoro-deoxy glucose PET–CT (positron emission tomography–computed tomography) revealed multiple avid nodules in bilateral lung fields predominantly in the periphery of lung fields (largest in left lung lower lobe measuring $2.5 \times 2.8 \times 2$ cm with speculated margins). Avid subcutaneous nodule was also seen in the left mons pubic region. Rest of the body showed physiological uptake (Fig. 3a and b). Complete blood counts, liver function tests and kidney function tests were all reported to be within normal laboratory limits. She was started on tab Letrozole 2.5 mg once daily. There was significant improvement of the vulvar lesion at 6 months of follow up (Fig. 1b). The satellite lesion has disappeared and the vulvar lesion has shown a decrease in erythema and indurations. A repeat PET–CT (Fig. 3c and d) also revealed decrease in the uptake and size of the lung nodules and significant reduction of the size of left lower lung lobe nodule (now measures 7 mm) and decrease in avidity of the subcutaneous vulvar lesion.

The patient is continuing Letrozole tablet and is on follow up now.

Discussion

Association of vulvar and breast cancer is very intriguing. Mammary gland at an ectopic site (along the primitive milk streak, extending from axilla to the groin) has been found to be a source of benign or malignant lesion of the breast. Associations of various types have been reported.

Primary breast cancer of the vulva is extremely rare [6]. Synchronous or metachronous association of infiltrating ductal carcinoma of breast and vulva has also been reported. Metastasis of breast cancer to vulva and vice versa is a very rare association. Vicus et al. reported metastasis of primary carcinoma of vulva to breast in a 49 year old female [7]. Primary carcinoma of breast with vulvar metastasis was first reported in 1964 by Convington et al. [8].

In cases of diagnosis of breast carcinoma of the vulvar tissue, distinction between primary breast carcinoma of vulva and vulvar metastasis from breast cancer needs to be distinguished. A prior history of breast cancer in the patient, identical histological and hormone receptor status of both breast and vulvar lesion and absence of in-situ elements may help in clinching the diagnosis of metastatic lesion [9]. Exact mechanism of this metastatic pattern is not clearly known, although vascular space involvement has been thought to be a probable cause. Altered lymphatic drainage after surgery may also be a cause of this pattern as it was reported by Valenzano et al. [2]. In this report, 49 year old patient developed a rectus abdominis myocutaneous flap metastasis (3 year after surgery)

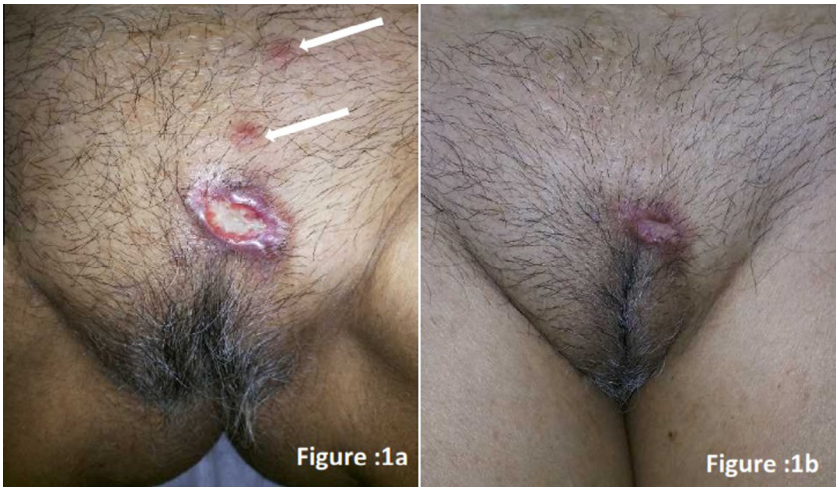

Figure 1 (a) Vulvar lesion with satellite nodules at presentation. (b) Same lesion after 6 months of hormone therapy.