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Case Report
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Coexistence of tuberculous axillary lymphadenitis and giant borderline malignant phyllodes tumor of the breast: A rare case report

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ABSTRACT

Cystosarcoma phyllodes are uncommon breast tumors which rarely metastasize to axillary lymph nodes. The tumor is similar to fibroadenoma in structure, but it is different histologically. Although surgery (excision vs. mastectomy) is the mainstay of treatment, the need for adjuvant therapies such as radiotherapy for a malignant variety is unclear. Its association with ipsilateral tubercular axillary lymph nodes has not been reported in literature so far. We report a 35-year-old female that presented with a giant borderline malignant phyllodes tumor of the right breast along with ipsilateral tubercular granulomatous axillary lymph nodes.

Key words: *Cystosarcoma phyllodes, axillary lymphadenitis, tuberculosis, mastectomy*

Introduction

Cystosarcoma phyllodes are primarily benign tumors and their name is ambiguous. They constitute 0.3 to 0.9% of all breast tumors, and the incidence of malignant phyllodes tumors is even lower. It was Müller who first recognized them in 1838. These tumors are classified as benign (60%), borderline (15%) and malignant (25%). This tumor appears exclusively in the female breast and nowhere else in the body [1]. Enlarged axillary lymph nodes in case of breast malignancy are not always due to metastasis. Mostly, these get enlarged to reactive hyperplasia or are rarely due to reactivation of dormant lying tubercular infection due to decreased resistance of the patient. Mycobacterium infection can affect a patient with malignancy, as these patients are immunologically compromised [2]. Thus, malignant phyllodes can present with granulomatous axillary lymph nodes compatible with tuberculosis. Such a case of borderline malignant phyllodes with tubercular granulomatous axillary lymph nodes is presented here.

Case Report

A 35-year-old female presented with a huge lump in her right breast. She narrated a history of small breast lumps for the last five years, which were treated with homeopathic medicine. In the last two months, there was a sudden increase in the present size. History of

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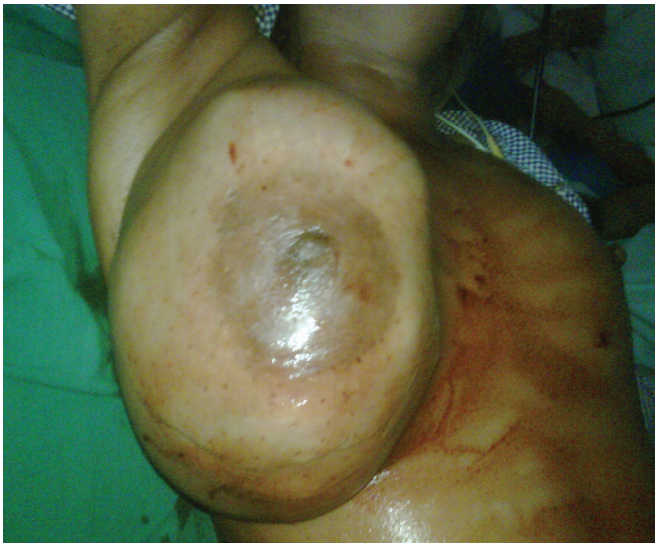

Figure 1. Showing the patient's breast photo.

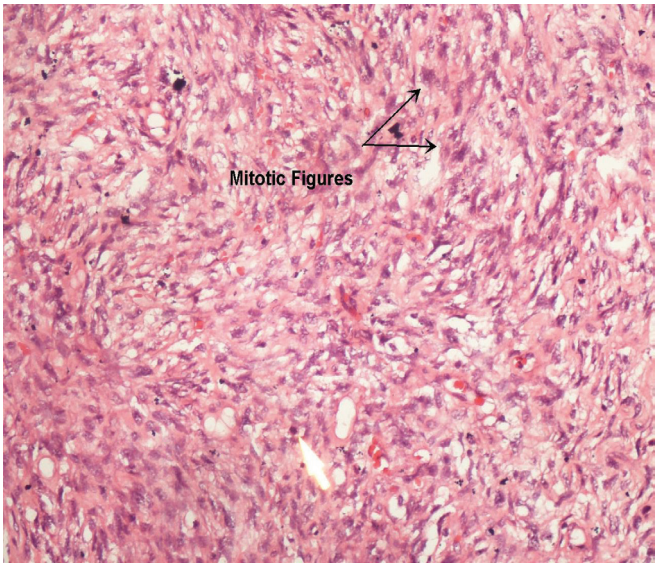

Figure 2. Photomicrograph of the same tumor. The stromal component is cellular with moderate atypia. Mitotic figures are seen (H&E, X200).

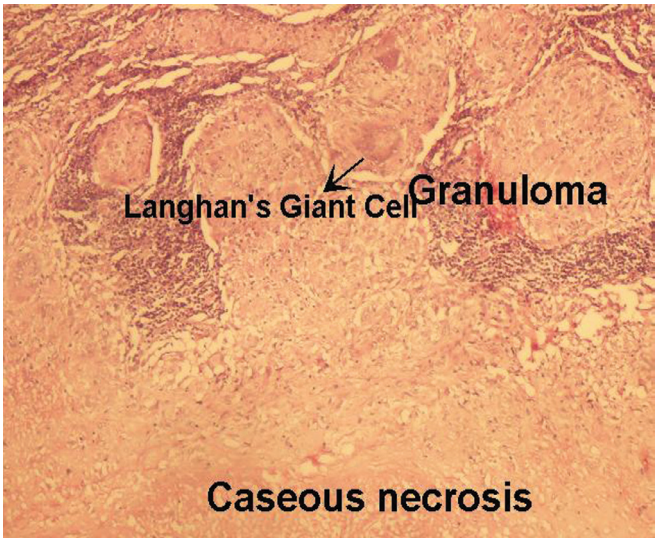

Figure 3. Photomicrograph of a section from the axillary lymph node showing granuloma with extensive caseation (H&E, X100).

slight pain in the right breast was also there. The patient was nulliparous, and there was no history of breast cancer in the family. On examination, the whole right breast was replaced by a tumor with a normal nipple and areola complex. The skin was stretched and glossy with dilated veins. It measured 24x18x10 cm, and was firm to cystic in nature. The skin was not adherent to mass, but the mass was adherent to underlying muscle of the chest wall (Figure 1). Ipsilateral axillary lymph nodes (medial group) were palpable, which were firm in consistency. The left breast and axilla were normal. An X-ray of the chest and ultrasonography of the abdomen were normal. FNAC of breast swelling revealed a borderline malignant phyllodes tumor. Right mastectomy with low axillary node clearance was done, with 21 lymph nodes being removed; the largest was measured at 2.5x1.5 cm and sent for histopathological examination. The main tumor's histopathology report revealed a borderline malignant phyllodes tumor (Figure 2) and that all of the lymph nodes revealed tubercular granuloma (Figure 3), which was an unexpected outcome. Thorough postoperative investigation of our patient did not reveal any evidence of pulmonary or other extrapulmonary tuberculosis. Since axillary node tuberculosis was not suspected preoperatively, a Mountex test, ESR and polymerase chain reaction (PCR) were not done. Postoperatively, the patient was put on antitubercular therapy (ATT). The patient was followed up to one year and there was no recurrence.

Discussion

Phyllodes tumors are fibroepithelial neoplasm. They make up 0.3 to 0.5% of female breast tumors and have an incidence of about 2.1 per million. The peak incidence of tumors occurs in women aged 45 to 49 years. The tumor is rarely found in adolescents and the elderly [3]. The majority of patients present with a macrolobulated mass that is firm, well defined, round, and painless, varying in size from 5cm to 30 cm. However, tumors bigger than 20 cm are called "GIANT TUMORS" (as in our case). Unusually, the left breast is more commonly involved than the right breast (the right breast was involved in our case) [1]. These tumors are classified as benign (60%), borderline (15%) and malignant (25%) [1]. Metastasis may occur either at the time of presentation or as late as 12 years after

[4], and mostly spreads by a hematogenous route to the lungs (66%), bones (28%), and brain (9%) — and rarely to the liver and heart [5]. Mostly, regional lymph node enlargement is reactive and rarely involved by tumors [4-6]. Axillary lymph node involvement ranged from 1.1% to 3.8% [6-9]. Approximately, 3 to 12% of malignant phyllodes tumors metastasize to lymph nodes [6,7]. Enlarged axillary nodes are not always due to metastasis. Mostly, they are reactive and may rarely be granulomatous. The occurrence of granulomatous necrosis in axillary lymph nodes can be a coincidence, the activation of dormant mycobacterium tubercular infection due to decreased immunity in malignancy, or may be either a retrograde spread from the mediastinal nodes or hematogenous spread from a sub-clinical focus, not picked by routine investigations in endemic areas like India [10]. An immune mechanism against infections, non-neoplastic and neoplastic conditions results in a granulomatous reaction in draining lymph nodes. A granulomatous reaction can also result due to infective agents such as mycobacteria, fungi, parasites, and brucellosis, and due to non-infective conditions such as sarcoidosis, foreign bodies, Wegener's granulomatosis, and traumatic fat necrosis. However, caseation is seen only in tuberculous inflammation (as in our case).

Primary breast cancer and axillary node tuberculosis can coexist, but phyllodes tumors are rarely malignant and their coexistence with axillary node tuberculosis has not been reported so far. Thorough investigation of our patient did not reveal any evidence of pulmonary or other extrapulmonary tuberculosis. Since axillary node tuberculosis was not suspected preoperatively, a Mountex test, ESR and polymerase chain reaction (PCR) were not done.

It is extremely difficult to differentiate a small phyllodes tumor from a fibroadenoma, which is sometimes treated with a conservative approach; these tumors then suddenly change to giant tumors, which may be a sign of malignancy (as in our case). For this reason, early diagnosis of the small phyllodes tumor is vital so that the correct management of the tumor, which often does include surgery, can be advised as early as possible. Usually, only in 10 to 20% of cases of a breast lump could a preoperative diagnosis of cystosarcoma phyllodes be made [8]. Cystosarcoma phyllodes can be histopathologically differentiated from a fibroadenoma breast by their cellular pattern, having increased atypical cellular changes and excessive stromal growth.

There is no consistent and reliable way to distinguish between small phyllodes tumors and other benign-appearing tumors on an ultrasound or mammography [9]. Other new diagnostic modalities are color Doppler ultrasound and magnetic resonance imaging (MRI). However, there are no fixed criteria which are accessible in mammography, ultrasonography (USG) and magnetic resonance imaging (MRI) to differentiate between cystosarcoma phyllodes and fibroadenoma [11].

Core tissue is better than Fine-needle aspiration cytology (FNAC) for the diagnosis of a phyllodes tumor [9,12].

There is no standard regimen of management of cystosarcoma phyllodes and it is still controversial. An adequate margin up to 1 cm of normal tissue along with the mass is excised. Wide local excision for small tumors and simple mastectomy for larger ones are usually satisfactory. If fascia or muscle is infiltrated (incidence of 2.4%), the excision of muscle is then done (as in our case). For tumors bigger than 5 cm, mastectomy with low axillary node dissection is performed if axillary nodes are also palpable [6], as in our case. Adjuvant radiotherapy is beneficial in a few cases of malignant and borderline cases of cystosarcoma phyllodes and also in tumor-recurrence cases (about 15 to 20%) [13]. The usefulness of cytotoxic drugs like anthracyclines, ifosfamide (Ifex), cisplastin, and etoposide has been unproven so far [14,15]. Hormone receptors like estrogen and progesterone have been documented in these tumors, but the use of hormone therapy like tamoxifen is still under investigation and further study is needed [16].

The recurrence rate is about 28–50% in literature. The most important risk factor for recurrence is the resection being done within 1–2cm negative surgical margins. Chen et al. have described how age, type of surgery, increased mitotic activity, and excessive stromal activity are usual risk factors for local recurrence [17].

Conclusion

Management of the phyllodes tumor presents the surgeon with unique challenges. Diagnostically, core tissue biopsy represents an important means for pre-

operative diagnosis, and helps in the differentiation of phyllodes tumors from fibroadenoma. The majority of these tumors can be managed by simple mastectomy. Axillary lymph node metastasis is rare. Mostly, axillary lymph node enlargement is reactive but may harbor metastatic despites of malignant phyllodes and/or can very rarely harbor dormant tuberculous granuloma, particularly in endemic countries like India. In such patients with palpable lymph nodes, low axillary nodes clearance is done along with mastectomy.

Though coexistence of phyllodes and tubercular axillary lymphadenitis is extremely rare, it should be kept in the back of the mind, particularly in endemic countries. Since the presence of tuberculosis may alter the postoperative management of the patient, a thorough investigation of tuberculosis is obligatory.

Conflict of interest statement

The authors have no conflicts of interest to declare.

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