

# Synchronous Bilateral Breast Benign Phyllodes Tumors in an Adolescent Female along with Depression: A Case Report

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## Abstract

Cystosarcoma phyllodes or phyllodes tumors are a rare neoplasm of the breast and present as painless breast mass. It accounts for 1% of all breast tumors. Whether benign, borderline or malignant, they have a high potential of recurrence. The bilateral occurrence of these biphasic tumors is rare. According to the literature, most of them are malignant and asynchronous. Synchronous bilateral benign presentation in a young nulliparous female is extremely rare. The definitive treatment of the benign entity is either wide local excision or mastectomy depending on size and age of the patient. We report a case of synchronous bilateral benign phyllodes tumors in a 22-year-old female associated with depressive features. Bilateral breast biopsy revealed benign phyllodes tumors. The case under report finds rarity due to its synchronous bilateral benign presentation in an unmarried adolescent female where the decision making regarding its management with surgical procedure becomes difficult and the significance of treating the associated depression in such a patient.

**Key words:** Adolescent female, Bilateral benign tumors, Breast, Depression, Phyllodes tumor, Synchronous

## INTRODUCTION

Phyllodes tumor is a rare fibroepithelial neoplasm of the breast in children and adolescents. In most of the cases, it presents as a rapidly growing, clinically benign breast mass. These tumors usually present as unilateral painless, well-circumscribed, mobile breast masses.<sup>1</sup> Bilateral tumors are rare entity and usually malignant and asynchronous in nature.<sup>2</sup> These tumors are locally aggressive and have high recurrence rate. Surgery is the mainstay of treatment.<sup>1</sup> We report a case of synchronous bilateral benign phyllodes tumors in a 22-year-old woman associated with depressive features. Local excision performed for the right side tumor and mastectomy with breast reconstruction performed for the left side tumor. There was an improvement in depressive features that was analyzed with Hamilton

depression rating scale (HDRS) with decrease in score after surgery and with appropriate pharmacotherapy.

## CASE REPORT

The 22 years young adolescent nulliparous female presented with bilateral breast lumps since 18 months. The patient developed features of depression during that period. Bilateral breast examination revealed lump on both the breasts without any axillary lymph node enlargement. Right breast examination showed a lump of size 6 cm × 6 cm, non-tender in nature, firm to hard in consistency, with lobulated appearance and normal intact nipple-areolar complex without any skin or chest wall fixity. Left breast examination revealed a lump of size 10 cm × 10 cm, non-tender in nature, firm to hard in consistency, with lobulated appearance and normal intact nipple-areolar complex without any skin or chest wall fixity. Contrast enhanced computed tomography scan of neck, chest, abdomen, and pelvis revealed lobulated well defined minimally enhancing bilateral breast masses (Figure 1). Careful examination ruled out other possible sites of lesion. Tru-cut biopsy from both the breast revealed

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features of benign phyllodes tumors (Figure 2). The patient was referred for psychiatric evaluation as at the time of presenting she was having low mood, crying spells and failed to carry out routine activities effortlessly. She was growing more aloof, and her HDRS score was calculated at first interview to assess the severity of the symptoms. The patient's score was HDRS 16 at the first interview. Surgery was performed on both the breast. A wide local excision with clear surgical margins was performed for right side benign phyllodes, whereas mastectomy with breast reconstruction was performed for left side benign phyllodes due to larger size of the tumor. Following surgery, the patient's general condition had markedly improved; she had gained appetite and weight. She was advised sertraline 50 mg at morning and clonazepam 0.25 mg once after dinner if required at the time of discharge. After 2 months of follow-up, the patient's HDRS score became drop to 3.

## DISCUSSION

Johannes Muller (1838) first used the term cystosarcoma phyllodes.<sup>3</sup> In 1982, the World Health Organization declared the "phyllodes tumor" as the most appropriate term among many synonyms. Phyllodes tumors account for

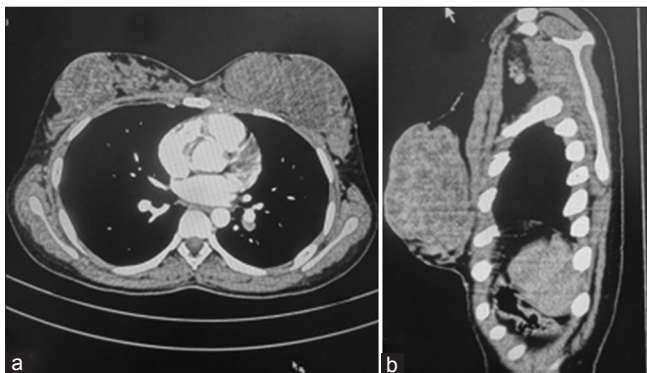
<0.5% of all breast malignancies.<sup>4</sup> The vast majority occur in women, in whom the median age at presentation is 42-45 (range 10-82 years).<sup>5</sup> This case is a 15 years female which is unusual. Synchronous bilateral multifocal phyllodes tumors are seen in a limited number of case reports.<sup>2</sup> Most of these reported have malignant form of the disease, at least, one side of the breast. The synchronous bilateral benign presentation is an extremely rare entity.

Phyllodes tumors are classified as benign, borderline and malignant basing on microscopic findings including stromal cellularity, cellular pleomorphism, mitotic activity, margins appearance, and stromal distribution.<sup>6</sup> Benign and malignant variants constitute 35-64% and 25%, respectively, and the rest are borderline.<sup>4</sup> All the subtypes of phyllodes tumors have a high propensity for recurrence. This case had synchronous bilateral presentation and met all the criteria for benign phyllodes tumor of both the sides.

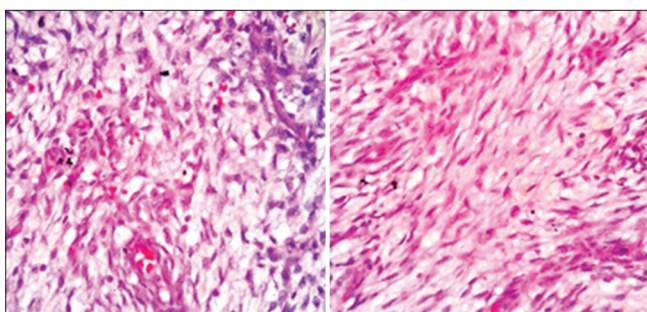
Phyllodes tumors are usually rapidly growing. The size varies from 1 to 41 cm with an average of 7 cm.<sup>4</sup> About 20% of the cases grow beyond 10 cm (giant phyllodes tumor) and rarely attain size of 40 cm when it is a unique challenge to the surgeon with respect to treatment options. Among children and adolescents, most phyllodes tumors exhibit a benign behavior.<sup>7</sup> The present case is a bilateral benign phyllodes tumor in an adolescent female. There are no standard etiologic or predisposing factors available for phyllodes tumors, except Li-Fraumeni syndrome, a rare autosomal dominant condition which is characterized by the development of multiple tumors.<sup>8</sup> Most bilateral phyllodes tumors in young women are associated with pregnancy or lactation.<sup>2</sup> Lactation is a precipitating factor in the metamorphosis of bilateral phyllodes tumor.<sup>2</sup> However, the role of female hormone still remains unclear. The present case is without pregnancy and lactation.

Phyllodes tumors present as a nonpalpable mass in 20% of cases and are identified on screening mammography.<sup>9</sup> Mammographically, phyllodes tumors are well defined with a smooth and occasionally lobulated border.<sup>10</sup> On ultrasound examination, phyllodes tumors often show smooth contours with low level homogeneous internal echoes and the absence of posterior acoustic enhancement.<sup>11</sup> There are no reliable mammographic or ultrasonic indicators available to differentiate between benign and malignant lesions.<sup>10,12</sup> Magnetic resonance imaging (MRI) provides more accurate extent of the disease prior to surgery, but, few data support the routine use of it.<sup>13</sup> MRI gives a better idea regarding the achievement of adequate margins when mastectomy to be performed.

Both phyllodes tumors and fibroadenomas are a spectrum of fibroepithelial lesions. It is difficult to distinguish



**Figure 1: (a and b) Contrast enhanced computed tomography scan of thorax showing bilateral minimally enhancing lobulated breast masses (left side mass [8 cm x 10 cm] larger than right side [6 cm x 6 cm])**



**Figure 2: Microsection shows presence of stromal hypercellularity with proliferation of stromal cells only with normal glandular elements (H and E, x400)**

diagnosis of fibroadenoma and phyllodes tumors by fine needle aspiration cytology and core biopsy.<sup>14</sup> An accurate pre-operative diagnosis needs imaging studies or biopsy, and the intraoperative frozen section. Microscopically, phyllodes tumors are characterized by epithelial lined cystic spaces with a hypercellular stromal projection. The stromal elements are a key component for differentiation of phyllodes tumor from fibroadenoma, and distinguishing between benign and malignant variants. An epithelial component with stromal components differentiates the phyllodes tumor from other stromal sarcomas.<sup>4</sup> There is no standard diagnostic protocol for pre-operative diagnosis of this rare disease. The present case on pre-operative tru-cut biopsy revealed findings consistent with bilateral benign phyllodes tumor of the breast.

Phyllodes tumor is best treated with wide excision when the size of the tumor is small. Surgical management is the mainstay of treatment in all types of phyllodes. Depending on the size of the breast, and size, location, and number of tumors; complete surgical excision with clear margins, mastectomy with or without breast reconstruction are the few treatment options for benign phyllodes tumors. Each subsequent recurrence converts tumor into a more aggressive one.<sup>15</sup> In the case of giant phyllodes tumor, excision with required margins is not possible. Mastectomy should be the treatment of choice in such patients including recurrent disease.<sup>16</sup> Like ipsilateral phyllodes tumor, surgery is the mainstay of treatment for bilateral presentation. Radical surgery does not have any survival advantage.<sup>17</sup> Mastectomy has better results than breast-conserving surgery in borderline and malignant cases.<sup>18</sup> There is controversy regarding the role of chemotherapy and radiotherapy.<sup>19</sup>

Phyllodes tumors have favorable prognosis with 5 years disease-free survival rates of 96%, 74% and 66% after surgery for benign, borderline and malignant phyllodes tumors, respectively.<sup>4</sup> There is no correlation about risk factors between tumor size and local recurrence.<sup>20</sup> Tumor size in relation to the breast is important for the extent of surgery and pathological resection margins. The most important factor for local control is excision with clear margins.<sup>18</sup> Wider margins (>1 cm) have the lowest risk of recurrence.<sup>21</sup> The possibility of local recurrence is 6-10% in benign, 25-30% in borderline and over 25% in malignant variants. Distant metastasis occurs in 3-13% of cases and lung is the most common site of distance metastasis.<sup>22</sup>

Depressive clinical features may develop in young women with breast phyllodes tumor. HDRS is the most widely used clinician-administered depression assessment scale.<sup>23</sup> It has 17 questions on mood and other symptoms which are scored. It consists of a set of 17 questions on depressed

mood, feelings of guilt, suicide, insomnia in early and middle of night, insomnia early in the morning, work and activities, retardation, agitation, anxiety psychic, anxiety somatic, somatic symptoms of gastrointestinal, general somatic, genital symptoms, hypochondrias, loss of weight, and insight which are given scores. This provides with an objective score which is done before and after treatment to quantify the improvement in depressive symptoms.

The present case is treated with surgery. Left side breast phyllodes treated with mastectomy and breast reconstruction due to larger size of the tumor, whereas wide local excision with clear surgical margins was performed for right side breast phyllodes. There was a significant improvement in depressive features.

## CONCLUSION

A differential diagnosis of phyllodes tumor should be considered in a rapidly growing but clinically benign breast lump, especially in young women. A thorough pre-operative diagnosis of this rare disease is important to determine the surgical approach.

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