Case Report

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Bilateral benign giant phyllodes tumor in an adolescent female: a rare case report

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ABSTRACT

Cystosarcoma phyllodes or phyllodes tumor are rare fibroepithelial neoplasm accounting for only about 1% of the breast lesions in children and adolescents. Whether benign, borderline or malignant, they have high potential of recurrence. These biphasic tumors are having very low rate of bilateral occurrence and in that event they tend to be asynchronous. The definitive treatment of the benign entity is either wide local excision or mastectomy depending upon size and age of the patient. Ours is a case of 16 years old, orphan low socio-economic status female having bilateral giant phyllodes tumors that were confirmed on Fine needle aspiration cytology of both breasts. Wide local excision of the tumor could be done on the right side, and patient was discharged on 10th post-operative day with advice to report after 6 weeks for possible surgery on the left enlarged breast. The histopathology report was consistent with the findings of benign phyllodes of the breast. Patient reported for the second surgery after 10 months of the first surgery and was subjected in wide local excision on the left side also. The case under report finds rarity due to its huge size, bilateral presentation in an unmarried adolescent female where the decision making regarding its definitive management becomes extremely difficult.

Keywords: Giant phyllodes, Bilateral, Benign phyllodes, Malignant phyllodes, Adolescent female

INTRODUCTION

Cystosarcoma phyllodes or phyllodes tumor are rare fibroepithelial neoplasm accounting for only about 1% of the breast lesions in children and adolescents. The median age of occurrence of the disease is 40-50 years, having an estimated incidence of about 2.1 per million. The tumor is rarely found in adolescents and elderly. 2.3

In 1982, World Health Organization declared the term "phyllodes tumor" as the most appropriate among more than 60 synonyms. Furthermore, World Health Organization sub-classified them histologically as benign, borderline or malignant. 4 Benign variants constitute 35-64% while malignant tumors comprise 25%, and the rest are borderline. 5-7 Whether benign, borderline or malignant, they have high potential of recurrence.

These biphasic tumors are having very low rate of bilateral occurrence and in that event they tend to be asynchronous. 8,9 The definitive treatment of the benign entity is either wide local excision or mastectomy depending upon size and age of the patient. 10,11

The case under report finds rarity due to its huge size, multiple tumors with bilateral presentation in an unmarried adolescent female where the decision making regarding its definitive management becomes extremely difficult.

CASE REPORT

A 16-year-old, an orphan low socio-economic status female of the remote village presented to the hospital with a history of progressive enlargement of the breast since the age of 13 years. The enlargement of the breast increased rapidly

in last 4-6 months. Sensing it the curse of some deity or unnatural force, the guardians of the female took her to a local tantric, who used some local medicine and hot iron rods on both the breasts of the female. Instead of getting any relief (expected decrease in size of the breast), the female suffered multiple ulcers in right breast. The guardians took the female to the local doctor in the dispensary, who in view of multiple non-healing ulcers in the breast referred her to the higher center. The age at menarche was 13 years, and there was no history of menstrual disturbances. On examination of the patient, except mild pallor nothing significant was detected on general physical examination and systemic examination. Local examination of the breast on the right side revealed multiple non-healing crusty ulcers. The size of the breast in cranio-caudal dimension was 56 cm by 44 cm. There were five lumps in the right breast ranging from 12 cm to 6 cm.

Left sided breast was 52 cm by 42 cm with uniform enlargement having firm consistency. Patient was admitted in the hospital and all the investigations for general anesthesia were done in addition to fine needle aspiration cytology (FNAC)of both breasts. FNAC both breast revealed bilateral benign phyllodes tumor with fibro epithelial hyperplasia, without any atypia.

The management plan was discussed with the attendants of the patient, including consent for mastectomy, which they gave with reluctance. Only wide local excision of the tumor could be done on the right side, the attendants refusing to give consent for mastectomy in spite of the fact that there was hardly any normal breast tissue in the operated breast. The patient was discharged on 10th post-operative day with advise reporting after 6 weeks for possible surgery on the left enlarged breast. The histopathology report was consistent with the findings of multiple benign phyllodes of the right breast. The patient lost the follow-up and reported 10 months after the first surgery. This time she had complained of further increase in size of the left breast and no recurrence on the right side. Again FNAC of the left breast was done to rule out malignant transformation of the earlier benign variant. The FNAC finding was consistent with benign phyllodes. After investigations for general anesthesia, patient was subjected to wide local excision of the tumor on the left side also as the attendant refused to give consent for mastectomy. Post-operative histopathological examination was consistent with findings of benign phyllodes of the breast. Patient was discharged on 10th post-operative day with advice to remain in regular follow-up. Patient remained in follow-up for 4 months (Figures 1-4).

DISCUSSION

Although today's phyllodes tumors find their description as a giant type of fibro adenoma as early as 1774 yet it was Chelius who described this tumor in 1827. ^{12,13} Furthermore, Johannes Muller (1838) was the first person to use the term cystosarcoma phyllodes. ¹⁴ It was believed

to be benign until 1943, when Cooper and Ackerman reported on the malignant biological potential of this tumor. In 1981 the World Health Organization adopted the term phyllodes tumor and as described by Rosen sub classified them histologically as benign, borderline, or malignant according to the features such as tumor margins, stromal overgrowth, tumor necrosis, cellular atypia, and number of mitosis per high power field.^{2,15} The majority of phyllodes tumors have been described as benign (35-64%), with the remainder divided between the borderline and malignant subtypes. The term phyllodes tumor represents a broad range of fibro epithelial diseases and presence



Figure 1: Bilateral giant phyllodes tumor in an adolescent.



Figure 2: Right breast with multiple ulcers.



Figure 3: Completion right breast surgery.



Figure 4: 6 months after first surgery.

of an epithelial component with stromal components differentiates the phyllodes tumor from other stromal sarcomas. 6,16,17

The median size of phyllodes tumor is around 4 cm. 20% grow larger than 10 cm (giant phyllodes tumor) and rarely attains size of 40 cm, when it is a unique challenge to the surgeon with respect to treatment options. Higher incidence of phyllodes tumor has been reported in people of Asian heritage.¹⁸

Liang⁷ reported rare occurrence of phyllodes tumor in adolescents and elderly. Whereas, Tse et al.¹⁹ reported that in children and adolescents, most phyllodes tumors exhibit a benign behavior. Although bilateral phyllodes and phyllodes tumors in adolescents are a rare phenomenon, yet they have been reported sporadically in the surgical literature.^{17,20} Marti and Hiotis²¹ claimed to report the first case of multiple bilateral phyllodes tumor in the breast of an adolescent female. Ours is a case of bilateral giant benign phyllodes tumor in an adolescent female belonging to very poor socio-economic status.

Although pre-operative diagnosis of this rare disease is important to determine the surgical approach, yet there is no standard diagnostic protocol. Mammography, ultrasonography, FNAC, and core biopsies all are being practiced from case to case and center to center basis. Many studies have reported false negative. ^{22,23} In our study we had put our reliance on the FNAC, pre-operatively that revealed findings consistent with bilateral benign phyllodes tumor of the breast.

Surgical management is the mainstay of treatment in all types of phyllodes. In benign phyllodes depending upon the size of the breast and tumor, location of the tumor, number of tumors; complete surgical excision with clinically clear margins, mastectomy with or without breast reconstruction are the fewer options. Since an excision with required margins is not possible in giant phyllodes. Hence mastectomy should be the treatment of choice in such patients including patients with recurrent disease. ^{10,11,24-26} In view of age of the patient and her marital status, we could do wide excision of the phyllodes tumors on both sides in a gap of 1 year between two surgeries.

The possibility of local recurrence is 6-10% in benign, 25-30% in borderline and over 25% in malignant variants. Lung and bone are affected by metastasis commonly. The overall 5 year survival in benign and malignant phyllodes reported in the surgical literature is 91% and 82% respectively. 29

We in our case had no recurrence on the right side that was operated 18 months back and on the left side after 6 months of surgery. Patient is still under regular follow-up.

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