## **Case Report**

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# Fibroadenoma in an operated case of malignant phyllodes tumour: a rare case report

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#### **ABSTRACT**

Phyllodes tumours are rare fibroepithelial lesions that account for less than 1% of breast neoplasms. Displaying a broad range of clinical and pathological behaviour, phyllodes tumours should be regarded as a spectrum of fibroepithelial neoplasms rather than a single disease entity. We would like to present a rare presentation of fibroadenoma over site of a previously operated case of malignant phyllodes tumour. A 36 year old female came to OPD with complaints of lump in both breasts since 4 months. She gave history of previous surgery, i.e., left breast lumpectomy for which HPE report was suggestive of malignant phyllodes tumour. On examination, 3x2 cm, solitary, firm to hard, non-tender lump was felt in the junction of the upper outer and inner quadrant just below the scar of previous surgery and was not freely mobile and appeared fixed to the scar. Patient underwent excision of the lump with frozen section and final HPE report turning out to be fibroadenoma. Though diagnosis of malignant phyllodes tumour is not difficult, diagnosis of low grade phyllodes tumour and distinction from fibroadenoma on FNA is difficult due to overlapping features between the two lesions. Surgery remains the mainstay of treatment for patients with cystosarcoma phyllodes, extent of surgery been controversial, and continues to evolve. Incidence of fibroadenoma occurring at the previous operated site of malignant phyllodes tumour is very rare with no known previous documentation of such a case.

Keywords: Breast tumours, Fibroadenoma, Malignant phyllodes

#### **INTRODUCTION**

Phyllodes tumours are rare fibroepithelial lesions that account for less than 1% of all breast neoplasms. <sup>1-3</sup> First described by Johannes Müller in 1838, he coined the term cystosarcoma phyllodes; a misleading description as the tumours are rarely cystic and the majority follow a benign clinical course. In total, more than 60 synonyms have been reported but today, the World Health Organisation regards phyllodes tumour as the most

appropriate nomenclature.<sup>4-6</sup> Displaying a broad range of clinical and pathological behaviour, phyllodes tumours should be regarded as a spectrum of fibroepithelial neoplasms rather than a single disease entity. At one extreme, malignant phyllodes tumours, if inadequately treated, have a propensity for rapid growth and metastatic spread. In contrast, benign phyllodes tumours on clinical, radiological, and cytological examination are often indistinguishable from fibroadenomas and can be cured by local surgery. With the non-operative management of

fibroadenomas widely adopted, the importance of phyllodes tumours today lies in the need to differentiate them from other benign breast lesions.

A Medline search of the English literature published since 1975 was performed using the medical subject heading "phyllodes tumour". Further articles were identified from the reference lists of papers reviewed. In view of the rarity of these tumours, most reported clinicopathological series are small, retrospective in nature, with limited long term follow up.<sup>7</sup>

In this case report, we would like to present a rare presentation of fibroadenoma over the site of a previously operated case of malignant phyllodes tumour.

#### **CASE REPORT**

A 36 year old female came to the OPD with complaints of lump in both breasts since 4 months. The lump was not associated with pain and there was no history of associated nipple discharge. No history of weight loss or loss of appetite.

She gave history of previous surgery, i.e., left breast lumpectomy one year ago at a private hospital. HPE report of the lump excised then was suggestive of malignant phyllodes tumour. She doesn't give history of any other co-morbidity.

Patient was incidentally nulliparous and was married since the past 4 years.

On examination, in the right breast, there was evidence of 4x3 cm, solitary, firm, non-tender, freely mobile lump in the upper outer quadrant.

In the left breast, there was evidence of 5cm radial, old, healed and healthy scar of previous lumpectomy, 4cm superior to the nipple-areola complex at the 12 o' clock position. A 3x2 cm, solitary, firm to hard, non-tender lump was felt in the junction of the upper outer and inner quadrant just below the scar. Lump was not freely mobile and appeared fixed to the scar of the previous surgery.

FNAC of both breast lumps was suggestive of fibroadenoma. Patient was posted for excision of bilateral breast lumps under general anaesthesia. A circumareolar incison was taken for both lump excision and the lump from the left breast was sent for frozen section. Frozen section confirmed the swelling was in fact fibroadenoma and decision to avoid modified radical mastectomy was taken. Post-operative period was uneventful and patient was discharged on post-operative day 10 following confirmation of fibroadenoma as the final HPE report.

### **DISCUSSION**

Fibroadenoma (FA) and phyllodes tumor (PT) are fibroepithelial breast tumors that are characterized by

proliferation of both stromal and epithelial cells. Fibroadenoma is the most common breast tumor in adolescent and young adult women with a peak incidence in the third decade of life. This tumor usually presents as a small painless, solitary mobile mass.8 Phyllodes tumor, also known as cystosarcoma phyllodes, occurs much less frequently and accounts for 2-3% of all fibroepithelial neoplasms.8 The mean age of patients with PT is approximately 45 years, which is approximately 15 years older than the median age of patients with FA. Phyllodes tumor is usually larger in size than FA, and in many cases there is often a history of rapid growth. Both PT and FA usually appear as opaque well circumscribed masses by ultrasound or mammography.1 Some cases of PT may show indistinct borders, cysts, and cleftlike spaces on ultrasound scans. In general, however, it is not possible to reliably distinguish FA and PT by clinical or radiologic examination. Although FA is an entirely benign tumor, PT encompasses a wide spectrum that includes benign tumors at one end and overtly malignant tumors at the other end. Phyllodes tumor is categorized as benign, borderline, or malignant based on stromal characteristics (i.e., cellular atypia, mitotic activity, stromal overgrowth) and tumor margins.9-11 Benign PT is characterized by well-defined margins, minimal atypia, and rare mitotic figures, whereas malignant PT shows infiltrative margins, stromal overgrowth, significant stromal atypia, and more than 10 mitoses per 10 highpower fields.9 Lesions exhibiting features in between benign and malignant PT are categorized as borderline

PT. Surgical treatment of these tumors differs in accordance with their variable natural behavior. Thus, whereas FA is simply shelled out of the breast, PT is excised with removal of an adequate rim of the surrounding breast tissue to obviate any chance of local recurrence.

Fine-needle aspiration (FNA) biopsy is often the first mode of investigation of both palpable and non-palpable breast masses. Therefore, preoperative categorization of fibroepithelial tumors by FNA is important for the selection of appropriate therapy.

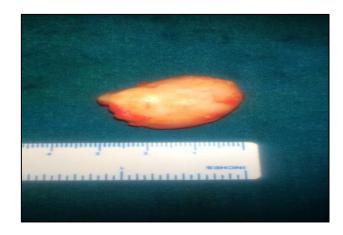


Figure 1: Excised specimen of fibroadenoma.

The diagnosis of malignant PT on FNA is usually straightforward and does not pose much of a problem. There is a striking predominance of stromal over epithelial component in the smears. Mitotically active, atypical and pleomorphic spindle cells constitute the hypercellular stromal fragments as well as the dispersed stromal cell population in the background. Though the diagnosis of malignant PT is not difficult, the diagnosis of low grade PT and its distinction from FA on FNA is difficult due to overlapping features between the two lesions. At the National Cancer Institute (NCI) sponsored conference for standardization of the reporting of breast aspirates, it was agreed on that fibroepithelial lesions with features suggestive of PT should be categorized as indeterminate. <sup>12</sup>



Figure 2: Post-operative photo showing previous operative site and new operative site.

Microscopically, phyllodes tumours are characterised by epithelial lined cystic spaces into which projects a hypercellular stroma (Figure 1). The presences of both epithelial and stromal elements are necessary to confirm the diagnosis. The stroma is the neoplastic component and determines the pathological behaviour. Only the stromal cells have the potential to metastasise. Primary tumours of the breast with pure sarcomatous differentiation and the absence of epithelial elements should be regarded as soft tissue sarcomas. Fibroadenomas are pathologically distinct from phyllodes tumours. They have a hypocellular stroma with few mitoses and little evidence of pleomorphism (Figure 2). Unlike phyllodes tumours, fibroadenomas have a true capsule.

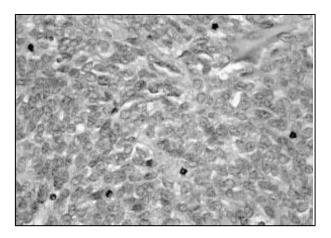


Figure 3: Malignant phyllodes tumour. Stroma shows cellular pleomorphism, nuclear atypia, and high mitotic activity (haematoxylin and eosin 200).

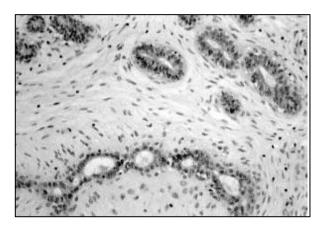


Figure 4: Fibroadenoma. Stroma shows no cellular pleomorphism, nuclear atypia, or mitotic activity (haematoxylin and eosin 200).

Cystosarcoma phyllodes is an uncommon tumor that can exhibit unpredictable behavior. Several histologic subtypes have been described using various classification systems. 9.18-20,22,27-29 However, these distinctions remain somewhat ambiguous and difficult to define. This is exemplified by the fact that although malignant tumors tend to be more aggressive both locally and distantly, metastases have been documented in patients with histologically benign tumors. 28,30

Although surgery remains the mainstay of treatment for patients with cystosarcoma phyllodes, the extent of surgical approach historically has been controversial, and continues to evolve. Local failure rates for benign tumors range between 5-15% and 20-30% for malignant tumors. 15,19,22,23,26,31-33 Margins of 1-2 cm have been recommended, and enucleation alone is insufficient. 19,21,22,25,26 de-Roos et al. found positive margins in all patients who sustained a local recurrence after local excision. 24 The reported rates of distant metastasis for patients with malignant tumors range from 25-40%. 9,19,25,31

The role of adjuvant radiotherapy is not well established. August et al suggested radiotherapy after wide local excision for high risk lesions (tumour >5 cms, presence of stromal overgrowth, >10 mitoses/10hpf, infiltrative margins), for local recurrence would likely be avoided in 90% or more of these patients.<sup>34,35</sup> The role of chemotherapy seems to be limited to the treatment of metastases and for palliation of unresectable local recurrences.<sup>36</sup>

Follow-up for malignant phyllodes tumour consists of biannual physical examination for 5 years, then annually; baseline unilateral mammogram 3 months post excision with or without radiotherapy should be done if breast was conserved, followed by annual bilateral screening mammography; biannual CT of chest to be performed for 2-5 years for high risk lesions.<sup>36</sup>

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#### **REFERENCES**

- 1. Dyer NH, Bridger JE, Taylor RS. Cystosarcoma phylloides. Br J Surg. 1966;53:450-5.
- Popescu I, Serbanescu M, Ivaschescu C. Phyllodes tumours of the breast. Zentbl Chir. 1991;116:327-36.
- 3. Buchanan ED. Cystosarcoma phyllodes and its surgical management. Am Surg. 1995;61:350-5.
- 4. Müller J. Uber den feineren Ban und Die Formen der Krankaften Geschwulste. Berlin: G Reiner. 1838:1:54-7.
- Fiks A. Cystosarcoma phyllodes of the mammary gland- Muller's tumor. Virchows Arch. 1982;392:1-
- 6. World Health Organisation. Histological typing of breast tumors. Tumori. 1982;68:181-98.
- Parker SJ, Harries SA. Phyllodes tumours. Postgrad Med J. 2001;77:428-35.
- 8. Rosen PP, Oberman HA. Fibroepithelial neoplasms. In: Atlas of tumor pathology tumors of the mammary gland. Washington, DC: Armed Forces Institute of Pathology; 1993.
- 9. Pietruska M, Barnes L. Cystosarcoma phyllodes: a clinicopathologic analysis of 42 cases. Cancer 1978;41:1974-83.
- Norris HJ, Taylor HB. Relationship of histologic features to behavior of cystosarcoma phyllodes: analysis of ninety-four cases. Cancer. 1967;20:2090-9.
- 11. Hart WR, Bauer RC, Oberman HA. Cystosarcoma phyllodes: a clinicopathologic study of twenty-six hypercellular periductal stromal tumors of the breast. Am J Clin Pathol. 1978;70:211-6.
- 12. The uniform approach to breast fine needle aspiration biopsy. A synopsis. Acta Cytol. 1996;40:1120-6.

- 13. Aranda FI, Laforga JB, Lopez JL. Phyllodes tumor of the breast. An immunohistochemical study of 28 cases with special attention to the role of myofibroblast. Pathol Res Pract. 1994;190:474-81.
- Fernandez BB, Hernandez FJ, Spindler W. Metastatic cystosarcoma phyllodes, a light and electron microscopic study. Cancer. 1976;37:1737-46.
- 15. Moore MP, Kinne DW. Breast sarcoma. Surg Clin North Am. 1996;76:383-92.
- 16. Blichert-Toft M, Hansen JPH, Hansen OH. Clinical course of cystosarcoma phyllodes related to histologic appearance. Surg Gynecol Obstet. 1975;140:929-32.
- 17. Bennett IC, Khan A, De Freitas R, Chaudary MA, Millis RR. Phyllodes tumours: a clinicopathological review of 30 cases. Aust N Z J Surg. 1992;62:628-33.
- 18. Kario K, Maeda S, Mizuno Y, Makino Y, Tankawa H, Kitazawa S. Phyllodes tumor of the breast: a clinicopathologic study of 34 cases. J Surg Oncol. 1990;45:46-51.
- 19. Reinfuss M, Mitus J, Duda K, Stelmach A, Rys J, Smolak K. The treatment and prognosis of patients with phyllodes tumor of the breast: an analysis of 170 cases. Cancer. 1996;77:910-6.
- 20. Cohn-Cedermark G, Rutqvist LE, Rosendahl I, Silfverswa d C. Prognostic factors in cystosarcoma phyllodes: a clinicopathologic study of 77 patients. Cancer. 1991;68:2017-22.
- 21. Modena S, Prati G, Mainente M, Massocco A, Montresor E, Pelosi G, et al. Phyllodes tumor of the breast: problems of differential diagnosis and therapeutic approach from an analysis of 17 cases. Eur J Surg Oncol. 1993;19:70-3.
- 22. Salvadori B, Cusumano F, Del Bo R, Delledone V, Grassi M, Rovini D, et al. Surgical treatment of phyllodes tumors of the breast. Cancer. 1989;63:2532-6.
- 23. Ciatto S, Bonardi R, Cataliotti L, Cardona G. Phyllodes tumor of the breast: a multicenter series of 59 cases. Eur J Surg Oncol. 1992;18:5454-9.
- 24. deRoos W, Kaye P, Dent D. Factors leading to local recurrence or death after surgical resection of phyllodes tumours of the breast. Br J Surg. 1999;86:396-9.
- 25. Rowell MD, Perry RR, Hsiu JG, Barranco SC. Phyllodes tumors. Am J Surg. 1993;165:376-9.
- Zurrida S, Bartoli C, Galimberti V, Squicciarini P, Delledonne V, Veronesi P, et al. Which therapy for unexpected phyllodes tumour of the breast? Eur J Cancer. 1992;28:654-7.
- 27. Norris HJ, Taylor HB. Relationship of histologic features to behavior of cystosarcoma phyllodes: analysis of 94 cases. Cancer. 1967;20:2090-9.
- Azzopardi J. Problems in breast pathology. Philadelphia: W.B. Saunders Company; 1979:355-79.
- 29. Reinfuss M, Mitus J, Smolak K, Stelmach A. Malignant phyllodes tumours of the breast. A

- clinical and pathological analysis of 55 cases. Eur J Cancer. 1993;29a:1252-6.
- 30. Mangi A, Smith B, Gadd M, Tanabe K, Ott M, Souba W. Surgical management of phyllodes tumors. Arch Surg. 1999;134:487-93.
- 31. Turalba CIC, El-Mahdl AM, Ladaga L. Fatal metastatic cystosarcoma phyllodes in an adolescent female: a case report and review of treatment approaches. J Surg Oncol. 1986;33:176-81.
- 32. Stebbing JF, Nash AG. Diagnosis and management of phyllodes tumour of the breast: experience of 33 cases at a specialist centre. Ann R Coll Surg Engl. 1995;77:181-4.
- 33. Staren ED, Lynch G, Boyle C, Witt TR, Bines SD. Malignant cystosarcoma phyllodes. Am J Surg. 1994;60:583-5.

- 34. August DA, Kearney T. Cystosarcoma phylloides: Mastectomy, lumpectomy or lumpectomy plus irradiation. Surg Oncol. 2000;9:49-52.
- 35. Harada S, Fujiwara H, Hisatsugu T, Sugihara H. Malignant cystosarcoma phyllodes with lymph node metastasis; case report. Jpn J Surg. 1987;17:174-7
- 36. Pandey M, Mathew A, Kattoor J Abraham EK, Mathew BS, Rajan B, Nair KM. Malignant phyllodes tumor. Breast J. 2001;7:411-6.

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