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Youngest case of ductal carcinoma in situ arising within a benign phyllodes tumour: A case report

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Highlights

- Phyllodes tumour is a rare type of breast tumour, accounting for less than 1% of benign and malignant breast tumours.
- Phyllodes tumour is classified pathologically as benign, borderline or malignant
- Phyllodes tumour with ductal carcinoma in situ can exist as separate foci or arise from within the tumour
- Phyllodes tumour usually presents in late 50s but has never been reported in a 23-year-old nulliparous woman.

Abstract

INTRODUCTION: Phyllodes tumour (PT) is a rare tumour of the female breast. The tumour clinically and radiologically mimics the features of a fibroadenoma. Ductal carcinoma in situ (DCIS) in the epithelial component of PT is a very rare finding.

PRESENTATION OF CASE: We present youngest ever case of a 23-year-old nulliparous woman with high-grade ductal carcinoma in situ arising within a benign phyllodes tumor. Macroscopically, it is a
homogeneous tumour with solid components. Microscopically, it features typical leaf-like pattern with hypercellular stroma with high-grade ductal carcinoma in situ.

DISCUSSION: To date, eight such rare cases of benign phyllodes tumour with ductal carcinoma in situ have been documented. We report the youngest case known in literature so far.

CONCLUSION: As this is a very rare presentation, it poses several challenges in regard to both management and follow-up.

Keywords

Phyllodes tumour, benign, ductal carcinoma in situ, young age.

Introduction

Phyllodes tumour is a rare type of breast tumour, accounting for less than 1% of benign and malignant breast tumours. Phyllodes tumour is classified as benign, borderline or malignant, with approximately 8–10% of Phyllodes tumours being malignant. Several histological parameters should be evaluated, including stromal cellularity, atypia, mitosis, stromal overgrowth, infiltrative borders and presence or absence of necrosis. The occurrence of Phyllodes tumour with breast cancer exhibits in two patterns: a separate coexistence within an ipsilateral or contralateral breast, or breast cancer occurring within the PT. The incidence of breast carcinoma in Phyllodes tumour accounts for only 1–2% of all Phyllodes tumours. To the best of our knowledge, only eight cases of benign phyllodes tumours with ductal carcinoma in situ have been reported in the literature.
We describe today the youngest reported case of ductal carcinoma in situ occurring within a benign phyllodes tumour in a 23-year-old nulliparous woman with no previous history of breast surgery or any family history of breast cancer.

**Case Report**

A 23-year-old nulliparous woman presented to us in our breast assessment clinic with a history of right breast lump, which had been present since 3–4 weeks, associated with discomfort and history of usage of oral hormonal contraceptive pills for 1–2 years. There was no family history of breast cancer or history of any breast problems. Clinically and radiologically, it was a 4-cm benign lump in the right upper outer quadrant, fairly mobile and not attached to any underlying structures or overlying skin. Axillary examination was normal. An ultrasound-guided core biopsy of this lump was done on an outpatient basis and was discussed thereafter in our multi-disciplinary meeting. The biopsy findings were suggestive of a fibroadenoma, with slight stromal hyper-cellularity raising the suspicion of a phyllodes tumour. The patient subsequently underwent a wide local excision of the lesion as a day case and was managed as per standard protocols during postoperative period.

On final histology, macroscopically the tumour was 50 x 40 x 35 mm in diameter and well circumscribed. On cutting, it was homogeneous with prevalent solid components. No necrosis or haemorrhage was identified. Microscopically, the tumour was hyper-cellular, and it was composed of a proliferation of monomorphic stromal cells forming a leaf-like structure. The mitotic index was very low (2 x 10 hpf), and no necrosis, atypia or pleomorphism was seen. (Fig.1&2)

The second component of the tumour was characterised by a proliferation of epithelial cells. The intraductal epithelial proliferation was arranged in a cribriform and solid pattern of growth; the tumour itself showed foci of comedo-type necrosis, pleomorphism and mitotic activity with nuclear hyperchromasia (Fig.2). The myoepithelial layer was unremarkable and was positive for p63. Overall, the
histopathological features were compatible with a biphasic tumour consisting of a benign phyllodes tumour with foci of high-grade DCIS without an invasive component (Fig 3) and appeared to be excised with narrow margins. She underwent further cavity shaves, and the final histology did not reveal any further DCIS or invasive disease.

**Discussion**

Phyllodes tumours are most common between the ages of 40 and 50 years and can sometimes present just like fibroadenoma. They have the tendency to grow rapidly and are broadly classified into three distinct groups:

a) Benign

b) Intermediate

c) Malignant

The epithelial component of phyllodes tumours (benign or malignant) may show a range of metaplastic (apocrine, squamous) and proliferative changes.

The age of the previously reported patients with coexistent carcinoma and phyllodes tumour ranged from 26 to 80 years, with most of the patients above 50 years. This case report presents the youngest case of benign phyllodes tumour with high-grade ductal carcinoma in situ.

Various authors have reported a variety of phyllodes tumour subtypes with invasive and in-situ component within or surrounding it. To the best of our knowledge, until the present case study, there had been only eight such reported cases of benign phyllodes with ductal carcinoma in situ. Our case is perhaps the first reported case in the series, presenting at 23 years of age. Leong et al, reported first ever case in 1980 of a 49-year-old woman having ductal carcinoma in situ arising in benign phyllodes tumour. Cole-Beuglet et al, described a case of benign phyllodes tumour with DCIS with LCIS in a 55-year-old.
Others, such as Knudsen et al. 5, Grove et al. 3, Ward et al. 4, Nio et al. 12, have also reported similar cases, but all these patients were in the age range of late 50s to 75 years.

**Conclusion**

There have been several cases reported in literature in the past about malignant phyllodes tumour with invasive breast cancer, and rarely, with benign phyllodes tumour. Our case represents one of those rare, isolated combinations of a high-grade ductal carcinoma in situ arising from within the benign phyllodes tumour, which has presented at a very young age. Understandably, this has given rise to many questions in regard to both further management and follow-up. In our multidisciplinary breast meeting it was agreed that this patient should have an index breast MRI followed by radiotherapy and annual mammography combined with clinical examination. This has also raised some concerns about the length of mammographic surveillance (radiation exposure) in this patient, as she is young and would be followed up until the age of 50 years.

**Conflict of interests:** None

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**Ethical Approval:** Not required

**Consent:** Consent from the patient has been obtained.

**Author Contributions:**

Sharat Chopra: Primary author and constructor of most of the report, review of literature, assistant operating surgeon
Vummiti Muralikrishnan: Operating surgeon, assisted with editing process for submission

Maurizio Brotto: Pathologist, provided histopathology information and slides

Guarantor: Sharat Chopra

References


Fig 1. H&E staining showing hypercellular stroma with absence of atypia or mitosis.
Fig. 2. H&E staining showing leaf-like structure in a monomorphic background

Fig. 3. H&E staining showing DCIS with comedo necrosis