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CYSTIC SQUAMOUS METAPLASIA IN A PHYLLODES TUMOUR.

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ABSTRACT

Phyllodes tumours are uncommon fibroepithelial lesions of the breast characterized by stromal proliferation and overgrowth. The epithelial component may exhibit hyperplastic and metaplastic changes, however, epithelial squamous metaplastic changes forming epidermal cysts are rare occurrences. We report a case of a 56-year-old woman presented with enlarging right breast mass with suspicious clinical and radiological findings. Clinically, she had a huge mobile mass in the right breast with a palpable axillary lymph node. Mammogram revealed a complex mass with BIRADS score 5. She underwent wide local excision. Histopathology revealed benign phyllodes tumour with multiple unremarkable epidermal cysts. Epidermal cysts arising from squamous metaplasia within a phyllodes tumour are rare. It may give rise to indefinite clinical and radiological findings with malignant transformation being a possibility.

Keywords: Breast, Epidermal cyst, Fibroepithelial lesions, Phyllodes tumour, Squamous metaplasia.

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Keywords: Breast, Epidermal cyst, Fibroepithelial lesions, Phyllodes tumour, Squamous metaplasia.

INTRODUCTION

Phyllodes tumours (PT) are uncommon fibroepithelial neoplasms of the breast characterised by stromal proliferation and overgrowth. They are further categorized as benign, borderline or malignant depending on histological features such as stromal cellularity, stromal atypia and mitotic activity.¹ The epithelial component within PT may exhibit a spectrum of metaplastic and hyperplastic changes.^{1,2,3} Epidermal cysts within a PT have been postulated to arise from squamous met-

aplasia of the ductal epithelium.⁴ It can give an impression of a complex lesion suspicious for malignancy on radiological imaging. Furthermore, reported cases of malignant transformation from epidermal cysts warrant careful examination of this seemingly benign entity.⁴ We report a case of a suspicious breast lump diagnosed as benign phyllodes tumour with multiple epidermal cysts arising from squamous metaplasia in a 56-year-old woman.

CASE REPORT

A 56-year-old woman with no background of medical illness was referred to our hospital for evaluation and management of the right breast lump. She noticed an enlarging palpa-

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ble mass in the right breast of 2 months' duration but otherwise was asymptomatic. She had no previous breast surgery, hormonal consumption or family history of breast cancer. On examination, she had a well-defined mobile mass in the right breast measuring approximately 18cm x 15cm involving mainly the upper quadrants and retroareolar region.

Mammogram revealed a well-defined high-density lobulated mass occupying the entire right breast with macrocalcification present within. Ultrasound showed a large heterogeneous hypoechoic mass occupying the entire right breast measuring approximately 13.0 x 11.9 x 5.4cm. There are scattered anechoic regions within the mass consistent with necrosis. An enlarged right axillary lymph node measuring 2.3cm x 0.8cm was also identified. Thus, BIRADS category 5 (suspicious for malignancy) was reported. The lump was diagnosed as fibroepithelial lesion favouring phyllodes tumour following trucut biopsy. She subsequently underwent wide local excision of the right breast lump without axillary lymphnode dissection.

Macroscopically, the excised mass measures 16 x 14 x 7.5cm. Cut sections showed a well-circumscribed tumour exhibiting whitish to pale yellowish cut surface with curved clefts, areas of necrosis and haemorrhage. There were also multiple cystic lesions measuring from 5 to 30mm in diameters within the mass with papillary cut surface and some containing cheesy material (Figure 1). The tumour was seen 1mm from the nearest surgical margin.

Histologically, the tumour is well-circumscribed and exhibits proliferation of breast stroma, ducts and acini (Figure 2). The hypercellular stroma form leaf-like pattern displaying areas of increased stromal cellularity interspersed with myxoid and hyalinized background. The stromal cells exhibit spindled to ovoid vesicular nuclei and conspicuous nu-

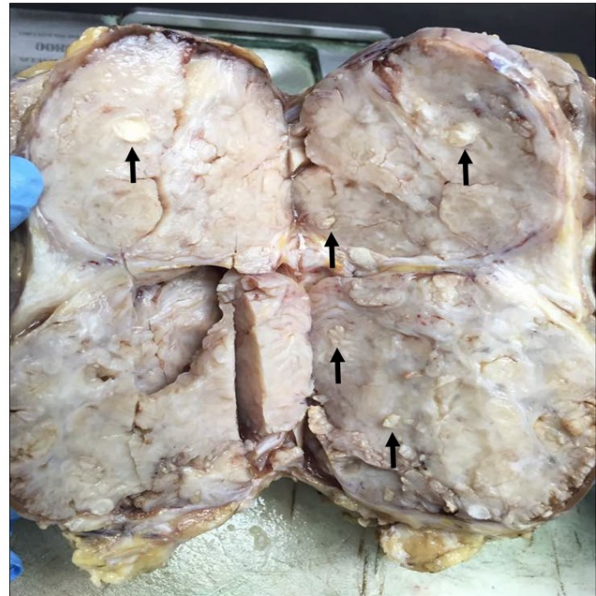


Figure 1: A well-circumscribed tumour with curved clefts and multiple cystic lesions containing white cheesy material (black arrows). (Click to enlarge image)

cleoli. Mitotic figures are observed at 0 to 2 per 10 high power fields. The ducts and acini are lined by two-tiered epithelial-myoepithelial cells and compressed by myxoid stroma into slit-like channels in some areas. Some of the acini display usual ductal hyperplasia, apocrine metaplastic epithelium and columnar cell change. Areas of necrosis and haemorrhage were noted. There was no calcification seen. Numerous epidermal cysts lined by stratified squamous epithelium containing lamellated keratin material, scattered blood vessels and plasma cells were present throughout the tumour. In areas, the stratified squamous epithelium was noted to arise from ductal epithelium. No heterologous elements were seen. The diagnosis of benign phyllodes tumour (PT) with multiple epidermal cysts was made.

The patient was followed up for 2 years and remained recurrence-free during the period.

DISCUSSION

Phyllodes tumours (PT) are uncommon fibroepithelial neoplasms of the breast charac-

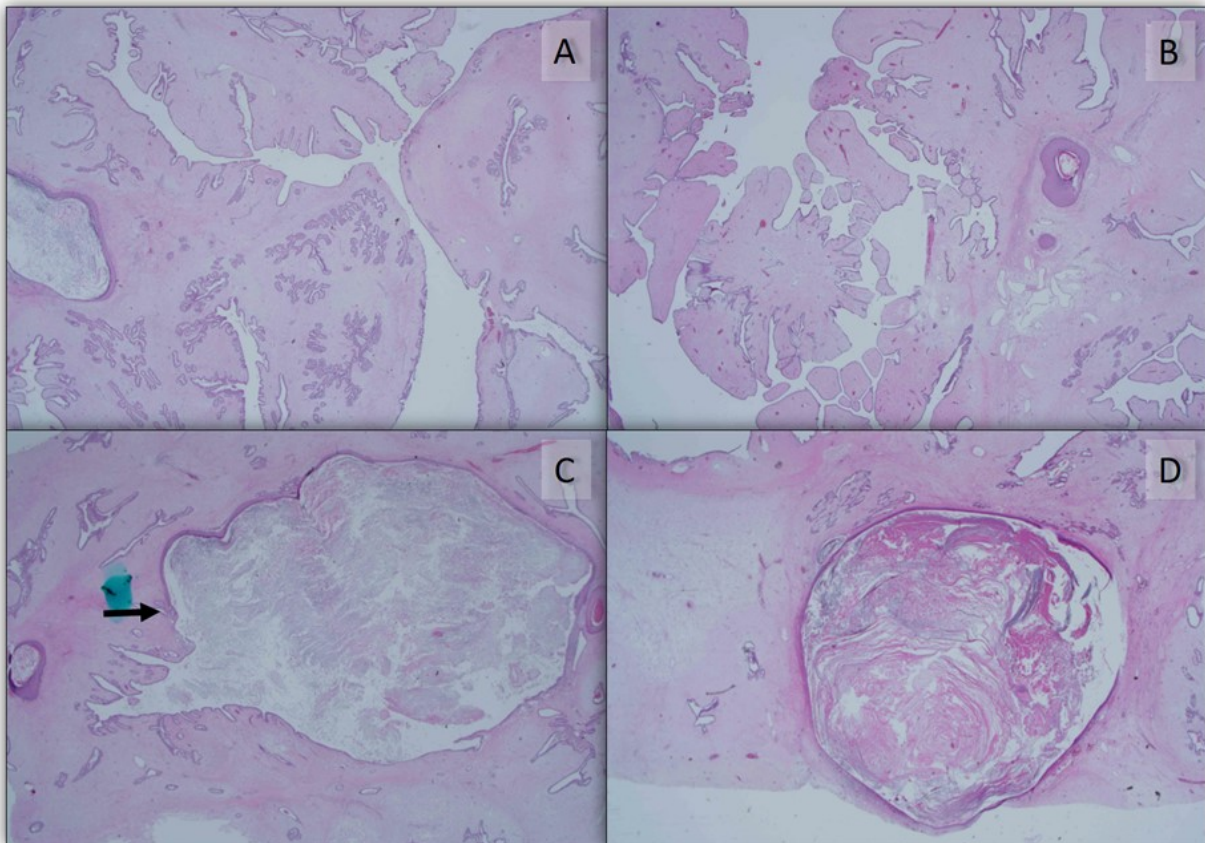


Figure 2: Phyllodes tumour exhibiting a leaf-like pattern of proliferating hypercellular stroma with scattered ducts and acini (A and B: H&E, 20x). Higher magnification showed numerous epidermal cysts present within the tumour (C and D: H&E, 40x). In areas, the stratified squamous epithelium is noted to arise from ductal epithelium (black arrow).

terised by stromal proliferation and overgrowth.¹ Patients usually present with solitary unilateral large breast mass that is mobile with smooth borders. Mammography and ultrasonography reveal a rounded or lobulated well-circumscribed mass that contain cysts or calcifications. The typical histopathological findings of PT are leaf-like architecture, increased cellularity of stromal component with presence of elongated two-layered epithelial-myoepithelial lined clefts that occasionally show cystic dilatation.^{1,2} Phyllodes tumour is further classified as benign, borderline or malignant depending on stromal cellularity, cellular atypia, mitotic activity count, stromal overgrowth and presence of heterologous component.¹

In our case, the tumour shows leaf-like pattern, mild cellularity and mitotic rate of less than 5 per 10 HPF, suggesting a be-

nign PT. The epithelial component may show a spectrum of proliferative changes from benign hyperplasia to squamous or apocrine metaplasia to ductal carcinoma.^{1,2} However, squamous metaplasia with formation of epidermal cysts within a PT as demonstrated in our case is a rare presentation with fewer than 15 cases reported. Previous published cases of epidermal cysts arising within phyllodes tumour are presented in [Table I](#). It is noted that epidermal cysts within phyllodes tumour has a wide age spectrum and the epidermal cysts can arise within any phyllodes tumour class, whether benign, borderline or malignant.

Epidermal cyst (EC) is a lamellated keratin-filled cyst lined by stratified squamous epithelium that is usually present within the dermis anywhere in the body.³ Solitary or pure epidermal cyst developing in the breast

Table I: Published cases of epidermal cysts arising from phyllodes tumour from 2015 to 2020.

Authors	Patient's age (years)	Size of phyllodes tumour	Histopathological features
Kumar et al., 2015. ⁵	55	Benign phyllodes tumour, 11 cm.	Multiple epidermal cysts, no other metaplastic elements.
Vasahar et al., 2015. ⁶	30	Benign phyllodes tumour, 12 cm.	Extensive squamous metaplasia with keratin cysts formation.
Gunadalaaimmanuel, 2016. ⁷	48	Benign phyllodes tumour, 17 cm.	Massive squamous metaplasia with keratin cyst formation.
Ugras et al., 2016. ⁸	59	Borderline phyllodes tumour, 4 cm.	Cystic spaces lined by multi-layered squamous cells filled with keratin material.
Chakrabarti et al., 2017. ⁹	41	Benign phyllodes tumour, 10 cm.	Squamous epithelium-lined cystic spaces filled with lamellated keratin.
Mukhopadhyay et al., 2017. ¹⁰	45	Malignant phyllodes tumour, 14 cm.	Extensive squamous metaplasia with formation of numerous epidermal cysts with chondrosarcomatous areas.
Muthusamay et al., 2018. ¹¹	38	Borderline phyllodes tumour arising from fibroadenoma, 10 cm.	Extensive squamous metaplasia of the duct epithelium and cystic spaces containing keratin flakes.
Rao et al., 2018. ¹²	65	Malignant phyllodes tumour, 9 cm.	Squamous metaplasia of duct epithelium with keratin cyst formation.

is predominantly found in the periareolar region and typically described as solid, well-circumscribed with complex or heterogeneous appearance on ultrasonography. Physical and imaging examination has been reported to simulate a malignant neoplasm.^{3,12,13} Formation of EC within the breast has been hypothesized to include obstructed hair follicles or injury to the epidermis that resulted in epidermal fragments implant and proliferate within the dermis. Pure epidermal cyst occurs without any preceding metaplastic changes of the native breast epithelium. However, EC may also arise from squamous metaplasia of normal breast columnar cells within a dilated duct of fibrocystic disease or phyllodes tumour³ as demonstrated in our case. EC arising in this setting may be termed cystic squamous metaplasia. It has been suggested the squamous metaplasia begin within the myoepithelial cell layer.⁸

Recognition of squamous metaplasia and EC is important due to documentation of malignant transformation to squamous cell carcinoma.³ Removal of PT with surgical margin of more than 1cm is recommended as width of margin less than 1cm is associated with increased recurrence rate.^{2,14} Local recurrence of PT has also been associated with

tumour size of more than 10cm and advance age of more than 50 years old, criteria which are present in our case.¹⁵ Axillary lymph node or distant metastases in a benign PT are rare² but the presence of enlarged axillary lymph node in this case may warrant further investigation and management to ensure malignancy is not missed.

CONCLUSION

Extensive epidermal cysts are a rare but important feature to recognize in a phyllodes tumour. They can simulate malignancy radiologically by having a complex appearance and may exhibit malignant transformation histologically. Adequate and extensive sampling for accurate diagnosis is essential. Presently, removal of phyllodes tumour containing epidermal cysts is the mainstay of treatment.

CONFLICT OF INTEREST

The authors have no conflicts of interest.

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