Benign phyllodes tumor of breast with extensive squamous metaplasia: A rare case entity

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ABSTRACT

Phyllodes tumor is a rare biphasic fibroepithelial neoplasm that comprises <1% of all neoplasms of the breast. A metaplastic changes in a phyllodes tumor, though reported, is an infrequently occurring event. Here, we present a case of a 30-year-old female who presented with a breast lump. Histopathological examination revealed a benign phyllodes tumor with florid squamous metaplasia. This case is presented for its rarity.

Key words: Phyllodes tumor, Squamous metaplasia, Keratin cyst

INTRODUCTION

Cystosarcoma phyllodes was the nomenclature given by Muller in 1838 to an uncommon bulky breast tumor with leaflike gross appearance (phyllodes - leaflike) having an aggressive clinical behavior. The World Health Organization (WHO) coined the term phyllodes tumor for the neoplasm originally called cystosarcoma phyllodes. The usage of this term is now considered inappropriate and the nomenclature suggested by the WHO is followed, phyllodes tumor comprises 0.3-1% of the neoplasm’s of breast. The median age of occurrence is around 45 years and is rare in younger age groups. These tumors can recur after an excision. Grossly the tumor resembles the giant fibroadenoma, but distinguished histologically from the later by more cellular connective tissue. They are locally aggressive and occasionally go for distant metastasis.

CASE REPORT

The 30-year-old female presented with complaints of a lump in her right breast for 6 months duration. On examination, a swelling was noted in the upper outer quadrant of the right breast. The swelling was firm, mobile and not tender. Nipple areola complex was normal. Ultrasonography breast suggested the features of fibroadenoma with degenerative changes. Clinical provisional diagnosis made as fibroadenoma and lumpectomy was done. The specimen was sent for histopathological examination.

Grossly, the lesion was a fairly circumscribed, nodular, growth measuring 12 cm × 10 cm × 6 cm. The cut surface was gray white in color with numerous solid and cystic areas. The cystic areas were filled with whitish chalky material.

Microscopy revealed a fibroepithelial lesion composed of a bilayer of epithelial cells within a cellular and myxoid stroma, projecting into the cystic spaces forming a leaf like a pattern (Figures 1a,b, 2,3). Areas of extensive squamous metaplasia with keratin cyst formation were present (Figures 4,5). The stromal cells had mild nuclear atypia, <5 mitosis/10 high-power field, no necrosis or infiltrative borders. Hence, the final diagnosis was given as “Benign phyllodes tumor with extensive squamous metaplasia.” The patient lost her follow-up.

DISCUSSION

Phyllodes tumor is a biphasic fibroepithelial neoplasm. It was first recognized in 1838 by Johannes
Figure 1 (a,b): Microscopic picture - the tumor shows a bilayer of epithelial cells within a cellular and myxoid stroma projecting into the cystic spaces forming a leaf-like pattern.

Figure 2: Microscopic picture - phyllodes tumor associated with fibroadenoma-like areas composed of compressed ductal elements and adjacent hypocellular myxoid stroma.

Figure 3: Microscopic picture - phyllodes tumor with areas of increased stromal cellularity and dilated cleft-like ductal elements admixed with areas of squamous metaplasia and keratin cyst (10x).

Figure 4: Microscopic picture - areas of squamous metaplasia associated with keratin cyst formation (x40).

Figure 5: Microscopic picture - keratin cyst filled with keratin material.

Muller. Phyllodes tumor constitutes 2.5% of all fibroepithelial lesions of the breast and 0.3-1% of all primary breast tumors. The tumor is common in people of certain ethnicity such as Hispanics. It occurs predominantly in middle-aged women and the nearly 15-20 years after the mean age of occurrence of fibroadenoma. Phyllodes tumor occur at a younger age group among Asians when compared to other parts of the world. Cases have also been reported in men. The time for development of a malignant phyllodes tumor is delayed by a period of 2-5 years when compared to benign phyllodes.\[1,3-5\]

Phyllodes tumor usually occurs as unilateral, painless, rapidly enlarging mass that rarely causes ulceration of the overlying skin of the breast. The size of the tumor is highly variable. Tumors may range in size from more than 10 cm to lesions <2 cm. They are well circumscribed, firm lesions showing surface bosselations. There is rarely any involvement of the overlying skin. The cut surface resembles a folded and compressed leaf bud showing a characteristic whorling pattern and cleft-like spaces. Cystic spaces, foci of hemorrhage and necrosis are found in some tumors.\[1,3,4\]

The cell of origin is the stromal cells in the intralobular or the periductal spaces. The tumor is biphasic composed of epithelium lined clefts and a cellular...
stoma. The epithelium is composed of two cell populations. The secretory epithelium and the myoepithelium. The epithelium may go in for changes such as hyperplasia. Cases of in situ carcinomas have been documented, but frank malignancies are rare.[1,3]

The tumor is graded as benign, borderline and malignant based on the stromal cellularity, overgrowth, nuclear atypia and mitotic count. Metaplastic changes in phyllodes tumor though reported is very rare. The common metaplasias known to occur are the apocrine and squamous metaplasia. In a study involving a series of phyllodes tumor of the breast, the stromal component underwent metaplastic changes forming adipose and chondromyxoid elements. Malignant heterologous components were seen in only 11 cases. Of the epithelial metaplastic changes squamous metaplasia occurred in 12 cases and among this only fewer cases showed keratin cyst, i.e., 5 cases.[1,3,5-7]

Squamous metaplastic changes in the breast resemble the lesions seen in the salivary glands and the female genital tract. It is believed to start in the myoepithelial cells and gradually involves the entire acinus. Immunohistochemical markers like actin, vimentin and S100 may be used to demonstrate the myoepithelial origin of the metaplastic squamous epithelium.[7]

Occurrence of keratin cyst in even rarer when compared to squamous metaplastic changes. Such a lesion in a fine needle aspiration may lead to a diagnostic confusion with dermoid cyst. The other lesions of breast that have been reported to present with squamous metaplastic changes include gynaecomastia and benign breast papillomatosis.[5]

We are here by documenting the rare finding of squamous metaplasia with keratin cyst formation in Phyllodes tumor. Squamous metaplasia in phyllodes tumor does not carry any clinical significance.

CONCLUSION
Phyllodes tumor of the breast is a rare fibroepithelial lesion. Metaplastic squamous changes in a phyllodes tumor with associated formation of numerous keratin cysts are a rare occurrence.

REFERENCES

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