Benign Phyllodes tumor with massive squamous metaplasia

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Abstract:
Phyllodes tumors are rare fibroepithelial tumors of the breast accounting for less than 0.5 percent of all breast neoplasms. Majority of these tumors occur in women, the median age at presentation is 42 to 45 years. They are classified by WHO into benign, borderline and malignant categories. Although squamous metaplasia is more often encountered in phyllodes tumors as compared to fibroadenomas, it is usually focal and not extensive. We present a case of benign phyllodes tumor with extensive squamous metaplasia and keratin filled cysts which has not been reported so far.

Keyword: Phyllodes, squamous metaplasia.

Figure 1: Benign Phyllodes tumor with leaf like pattern (H&E, 20x).

Case report:
A 48 year old female presented with recurrent swellings in her right breast for 4 years since 2009. She was evaluated at another medical center and underwent excision biopsy for the same. There was recent rapid increase in size. She also complained of associated pain over the swelling. There was no history of nipple discharge or deformity. She had no co-morbid illnesses. There was no family history of breast malignancy.

On physical examination:
Superficial dilated veins were present over the swelling. A large firm mass involving the entire right breast was present with areas of induration at 12 'o' clock position. Nipple was normal. There were no axillary lymph nodes palpable.

Ultrasoundography: Well-defined hypoechoic lesion measuring 17x5cm in right breast with posterior acoustic enhancement. No microcalcification/macronodularity.
Probably benign lesion-fibroadenoma/phyllodes BIRADS 3.

Operative findings: Right simple mastectomy under general anesthesia on 16.04.2013. The tumor was 16 x 10 cm and the tumor did not have infiltrative margins.

Gross appearance: The entire breast was found to be replaced by a multinodular mass measuring 17x10x5 cm with grey white to tan cut surface with cystic areas containing pultaceous creamish white material.

Microscopically a circumscribed tumor was seen composed of proliferated and compressed ducts separated by a cellular stroma composed of spindle-shaped cells displaying mild nuclear atypia. There was massive squamous metaplasia with keratin cyst formation. Areas of adenosis are present. Focally there was increase in stromal cellularity with mitotic figures, 1-2/10 hpf.

Discussion: Phyllodes tumors are rare fibroepithelial tumors of the breast accounting for less than 0.5% of all breast neoplasms(1,2). This lesion was first described by Johannes Müller in 1838 as “cystosarcoma phyllodes” due to it’s leaf like appearance and fleshy cut surface(3). The term phyllodes tumor was adopted by the WHO and is divided into benign, borderline and malignant categories. Clinical presentation: Majority of these tumors occur in women, ranging from 10 to 86 years, the median age at presentation is 42 to 45 years. These tumors are rare in women younger than 30 years of age. Studies have shown that the grade of these tumors increases with age at diagnosis(4). Some of these tumors have also been described in males. There are no known etiologic or predisposing factors associated with these tumors except for Li Fraumeni syndrome, which is a rare autosomal dominant condition characterized by the development of multiple tumors(5). On physical examination, the patients usually present with a smooth, multinodular well defined firm to hard discrete palpable tumor that is mobile and painless(6). The tumor varies from 1 to 41cm (average size of 4-7cm). Malignant phyllodes tend to be larger than benign variants, but there are exceptions to these.

Figure 2: Benign Phyllodes tumor with keratin filled cysts (H&E, 20x)
The histologic diagnosis was given as benign phyllodes tumor with massive squamous metaplasia. Immunohistochemically p63 positive myoepithelial cells were present around the ducts. Spindle cells were negative for HMWCK and CAM 5.2.
Figure 3: Benign Phyllodes tumor with squamous metaplasia and keratin filled cysts (H&E, 20x).

They usually occur as solitary unilateral masses. Rarely multifocality in a single breast or both breasts has been described.

Pathology:
Grossly they are round to oval well circumscribed unencapsulated multinodular masses with a firm bulging greyish white cut surface. Malignant tumors usually display foci of degeneration, necrosis and haemorrhage. Cystic areas containing keratotic debris are extremely rare. Microscopically the tumor arises from periductal stroma and contains scattered lobular elements. They demonstrate a heterogeneous histologic appearance. They are classified into benign, borderline and malignant categories. The criteria used to distinguish them into these categories take into account 1) Degree of stromal cellular atypia 2) Mitotic activity 3) Circumscribed or infiltrative margins and 4) presence or absence of stromal overgrowth. Benign phyllodes tumors have increased stromal cellularity, mild to moderate cellular atypia, circumscribed borders and less than 4 mitosis/10hpf and lack stromal overgrowth. Borderline phyllodes tumors have greater degree of stromal cellularity and nuclear atypia. Mitotic activity ranges from 4 to 9/10hpf. They usually have infiltrative borders and lack stromal overgrowth. Malignant phyllodes tumors demonstrate marked stromal cellularity and nuclear atypia with infiltrative borders and >10 mitosis per 10 hpf. They are characterized by the presence of stromal overgrowth. Squamous metaplasia of the ductal epithelium is seen in both benign as well as malignant phyllodes tumors in about 10% of cases. The presence of keratin filled cyst in phyllodes tumors is extremely rare. In FNAC, aspiration of cystic area of squamous metaplasia can lead to a wrong diagnosis of squamous cyst. A benign phyllodes tumor with extensive squamous metaplasia and keratin filled cysts has not been reported so far. The origin of squamous epithelium in the breast is from the myoepithelial cells. Squamous metaplasia occurs in a variety of benign and malignant breast lesions. Some of the benign lesions include fibroepithelial tumors, intraduct papilloma, duct ectasia, subareolar abscess and inflamed breast cysts. The malignant lesions include metaplastic carcinomas, adenosquamous carcinomas and squamous carcinomas. In phyllodes tumors squamous metaplasia is usually focal and not extensive. Squamous metaplasia is more often encountered in phyllodes as compared to fibroadenomas. The presence of squamous metaplastic cells in FNAC of suspected fibroepithelial neoplasms usually is towards phyllodes tumors.
Bibliography


