

**Case Report**  
**Epidermoid Metaplasia with Keratin Cyst Formation in**  
**Fibrocystic Disease of the Breast**

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**ABSTRACT.** A case of fibrocystic disease of the breast with massive epidermoid metaplasia is reported here. Squamous or epidermoid metaplasia is extremely rare in benign lesions of the breast and review of literature disclosed no other case reports of identical lesions in fibrocystic disease.

**Keywords :** Epidermoid Metaplasia — Keratin Cyst — Fibrocystic Disease

Despite its development as a cutaneous apocrine gland, the breast less frequently shows squamous or epidermoid metaplasia. While the squamous metaplasia is reported to occur in about one percent of mammary carcinomas<sup>1)</sup>, it is quite rare in benign tumors or non-neoplastic states of the breast<sup>2-6)</sup>.

Recently, we have encountered a case of fibrocystic disease of the breast in which massive epidermoid metaplasia of the ducts with keratin cyst formation was present. No reports of lesions identical to ours are found in the literature. Herein, the first case of epidermoid metaplasia in fibrocystic disease of the breast is presented.

**CASE REPORT**

A 35-year-old woman (Gravida 3, Para 3) noticed a mass in the left breast two months prior to a visit to the Kawasaki Medical School Hospital. Physical examination revealed multiple rubbery nodules in the lower outer quadrant of the left breast. Each nodules measured up to 0.8 cm in diameter and had smooth surface and ill-defined margin. No regional lymphadenopathy was present. Mammography was essentially benign with coarse and fine calcifications in places. Under the clinical diagnosis of fibrocystic disease, the lower outer quadrant of the left breast was widely excised (quadrantectomy).

**PATHOLOGICAL FINDINGS**

The resected specimen measured 7×7×2 cm. Cut surfaces showed gray

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fibrous tissue with yellow fatty areas scattered within it. The yellowish white lesions, measuring less than 2.5 mm in diameter, were seen within the fibrous areas. They tended to cluster in areas and was felt rubbery and solid.

Microscopically, the specimen consisted of mammary gland in fibroadipose tissue. The papillomatosis, adenosis, and cyst formation were seen in the ducts (Fig. 1). Cystic spaces were noted in the lobules. Although stratified squamous, apocrine and ordinary duct epithelia were present, the majority of the cysts were lined by stratified squamous epithelium with granular and horny layer, reminiscent of epidermis (Fig. 1, arrow). In some areas, the transition between metaplastic squamous epithelia and ordinary duct and/or papillomatous epithelia was seen (Fig. 1). Lumina were filled with lamellated keratin substances, which were partially calcified and even ossified. Frequently keratin cysts had been ruptured and associated with foreign body reaction (Fig. 2). No skin appendages were present near the cysts. Adjacent stroma was fibrous and a few lymphocytic infiltrates were seen near both ruptured and intact keratin cysts. Breast tissue nearby showed typical changes of fibrocystic disease. There was no evidence of cytological malignancy in either component.

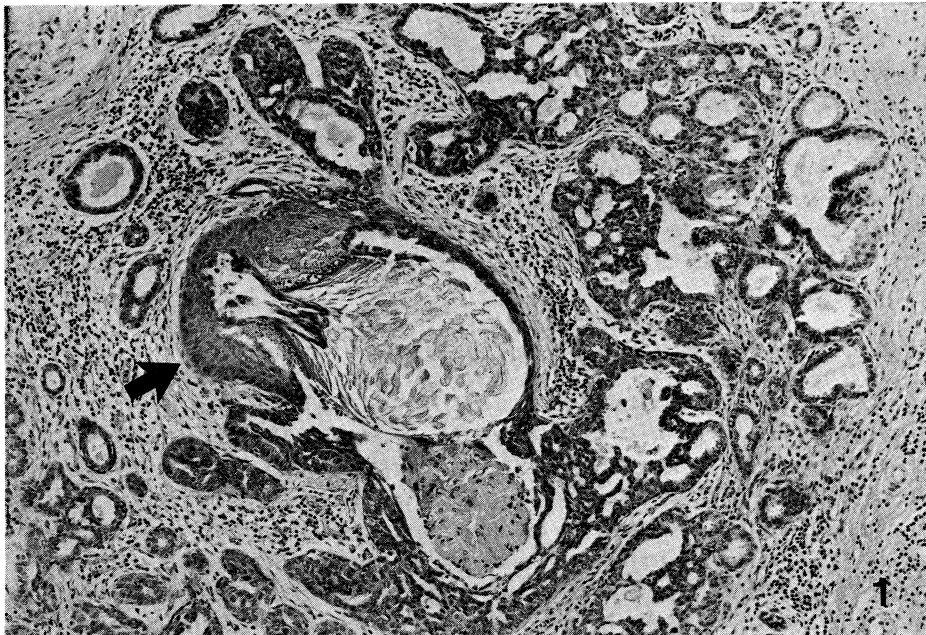


Fig. 1. Photomicrograph showing keratin cyst in the center (arrow). Note a gradual transition between papillomatous and squamous epithelium. (H-E,  $\times 150$ )

#### DISCUSSION

Except for the lactiferous ducts, no squamous epithelium is normally

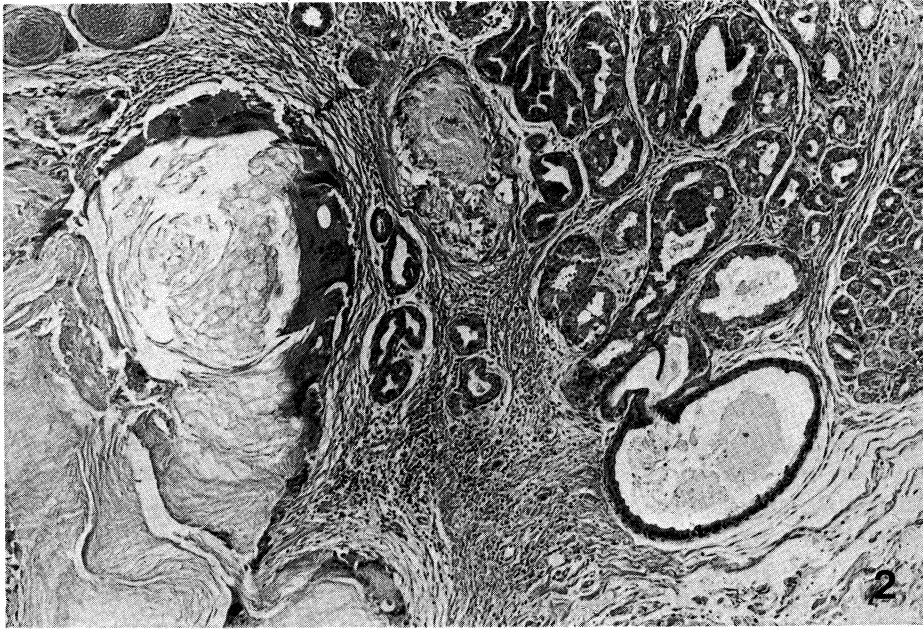


Fig. 2. Ruptured keratin cysts with foreign body reaction. Apocrine metaplasia is seen in adjacent ducts. (H-E,  $\times 150$ )

present in mammary glands. Occasionally, however, a squamous differentiation has been recognized in various abnormal conditions of the mammary gland. It is not uncommon in adenocarcinomas of the breast<sup>7)</sup>, while it is rather extremely rare in benign neoplastic and non-neoplastic states<sup>2-6)</sup>.

Simple dermoid cysts are known to occur in the breast<sup>8)</sup>. Congenital misplacement, implantation of epidermal fragments by trauma, and invagination of the skin epithelium have been offered to explain its existence within the breast. The association of sebaceous glands with dermoid cysts is frequently noted and seems to be a prerequisite for the diagnosis, thereby excluding our case from this category.

The squamous or epidermoid metaplasia results from the transformation of columnar or transitional epithelial cells and develops in a variety of organs, and of course, the mammary gland is not an exception. The gradual change from normal duct and/or papillomatous epithelia to the squamous in the case under discussion suggests its origin through metaplasia. Epidermoid metaplasia has been noted in benign tumors such as fibroadenomas<sup>2,3)</sup> or cystosarcoma phyllodes<sup>4,5)</sup>. Extrinsic factors may take part in this condition. Repeated injections of scarlet red and oleic acid or turpentine can induce squamous metaplasia in rabbit mammary gland<sup>6)</sup>. In addition, depletion of vitamin A has been shown to produce squamous metaplasia in respiratory tract, salivary gland, pancreas and prostate<sup>9,10)</sup>. It seems, however, that these factors are not

relevant to our case. Also, in animal experiment, Bonser<sup>11)</sup> has shown that an overabundant supply of estrogens may promote squamous metaplasia and keratinization, but this appears to have no counterpart in human physiology. Chronic inflammatory stimuli are the most common inducers of squamous metaplasia. Squamous metaplasia of the respiratory mucosa in smokers and patients with chronic bronchitis and that of bladder mucosa with stones are the example. Analogies may be sought in the breast. In fact, Willis<sup>6)</sup> reported epidermoid metaplasia in achronically inflamed mammary duct. In our case, nothing is known about the possible etiological factors except for the association of fibrocystic disease. Apart from the exact nature of fibrocystic disease of the breast, this disease per se or in combination with other factors may exert on for epidermoid change. Stasis of secretions within the mammary ducts are frequently encountered without squamous metaplasia. In contrast to the frequency of the disease in elderly woman, however, the association of squamous metaplasia and fibrocystic disease has not been reported to date. These together imply that squamous metaplasia in fibrocystic disease in our case may be merely a chance occurrence and some other factors must have existed. The etiology of squamous metaplasia in our case is entirely unclear and the discussion to this end is beyond the scope of the current communication. At any rate, our case is the first to present epidermoid metaplasia in fibrocystic disease of the breast.

#### REFERENCES

- 1) Willis, R.A. : Epithelial tumors of the breast. In *Pathology of the tumors*, 4th ed. London, Butterworth. 1960, pp. 208-256
- 2) Salm, R. : Epidermoid metaplasia in mammary fibroadenoma with formation of keratin cysts. *J. Path. Bact.* **74** : 221-222, 1957
- 3) Salm, R. : Massive epidermoid metaplasia with keratin cyst formation in a giant fibroadenoma of breast. *J. Path. Bact.* **77** : 297-299, 1959
- 4) Treves, N. and Sunderland, D.A. : Cystosarcoma phyllodes of the breast; A malignant and a benign tumor. *Cancer* **4** : 1286-1332, 1951
- 5) Norris, H.J. and Talor, H.B. : Relationship of histologic features to behavior of cystosarcoma phyllodes. *Cancer* **20** : 2090-2099, 1967
- 6) Willis, R.A. : Metaplasia. In *the Borderland of embryology and pathology*, 2nd ed. London, Butterworths. 1962, pp. 519-583
- 7) Cornog, J.L., Mobini, J., Steiger, E. and Enterline, H.T. : Squamous carcinoma of the breast. *A. J. C. P.* **55** : 410-417, 1971
- 8) Menville, J.G. : Simple dermoid cysts of the breast. *Ann. Surg.* **103** : 49-56, 1936
- 9) Blackfan, K.D. and Wolbach, S.B. : Vitamin A deficiency in infants. *J. Pediat.* **3** : 679-706, 1933
- 10) Editorial : Vitamin A and cancer. *Lancet* **1** : 575-576, 1980
- 11) Bonser, G.M. : Microscopical study of evolution of mouse mammary cancer : Effect of milk factor and comparison with human disease. *J. Path. Bact.* **57** : 413-422, 1945