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□ Case Report □

Aberrant Breast Tissue of the Perineum

—A Report on Two Cases—

Young Nyun Park[1], Hyeon Joo Jeong[1] and Kook Lee[2]

In this abstract we report on two cases of aberrant breast tissue of the perineum in a 41-year-old and a 42-year-old woman with the complaint of a slowly growing vulvar mass. The masses were not fixed, they were ovoid, rubbery firm and measured 3×2.5 and 4×3cm in size. Microscopically, they revealed normal lobular architecture with focal papillomatosis in the former.

Key Words: Breast, ectopic, perineum

Aberrant breast tissue may occur anywhere along the embryonic milk line which runs from the anterior axillary folds, to the inner thigh; examples are more comon above the umbilicus than below. The occurrence of breast tissue within the perineum is extremely rare. Deaver and Macfarland (1917) noted only 2 vulvar breast tissues in the review of nearly 11,000 examples of supernumerary breast tissue. Histologically, it is composed of breast tissue and it is subject to physiologic and pathologic changes including swelling (Two and Shanmugaratnan 1962; O'Hara and Page 1985), fibroadenoma (Burger and Marcuse 1954) or carcinoma (Cho *et al.* 1985).

We experienced two cases of ectopic breast tissue of the perineum and report them with a review of the literature.

CASE REPORTS

Casse 1

The patient, a 41-year-old female, was admitted due to a slowly growing, non-tender mass of the perineum for 6 months.

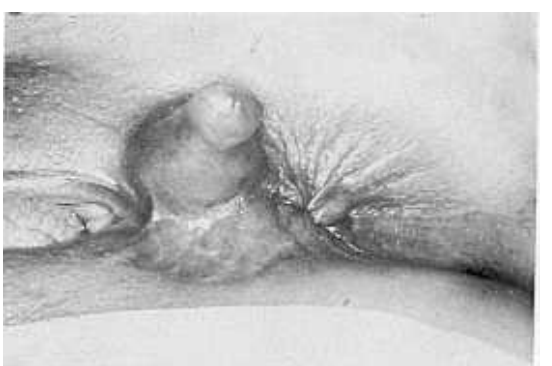

Fig. 1. *An ovoid fungating mass, measuring 3×2.5cm is located between the left labia majora and anus.*

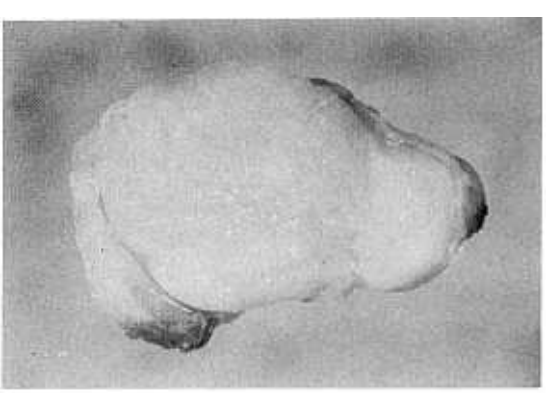

Fig. 2. *The cut surface of the mass is a pinkish tan and slightly lobulated.*

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Department of Pathology[1], Yonsei University College of Medicine, Seoul, Korea
Department of Obstetrics and Gynecology[2], Yongdong Severance Hospital, Yonsei University College of Medicine, Seoul, Korea
Address reprint requests to Dr. Y N Park, Department of Pathology, Yonsei University College of Medicine, C.P.O. Box 8044, Seoul, Korea

Her obstertrical history was gravida III, para II, living II, death 0, abortion I and her last delivery was 2 years ago. On physical examination, the fungating mass was located between the labia majora and the anus, and was movable (Fig. 1).

Grossly, the enucleated mass, measuring 3cm in diameter, was roughly ovoid and rubbery firm. On cut section it showed a pinkish tan, slightly lobulated appearance (Fig. 2). The microscopic findings were those of the normal breast with focal papillomatosis. There was neither cytological atypia nor malignant change (Fig. 3).

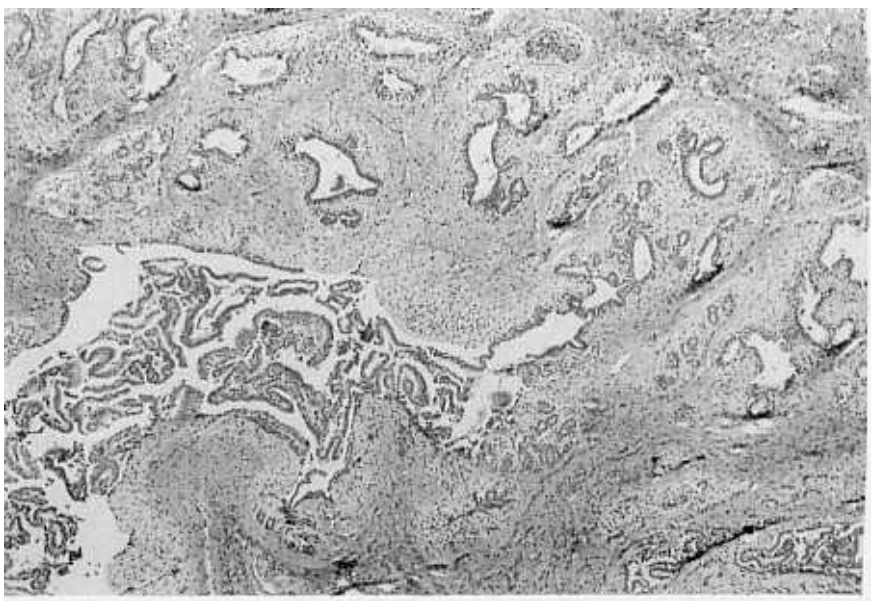

Fig. 3. Breast tissue with focal papillomatosis H-E, ×40.

Fig. 4. Breast tissue H-E, ×40.

Case 2

The patient was a 42-year-old female with a slowly growing vulvar mass. Grossly, the mass was solid, rubbery and measured 4×3×3cm. Cut sections revealed an encapsulated mass with a smooth, homogenous grayish-white surface. The microscopic findings were those of the normal breast (Fig. 4).

DISCUSSION

Mammary tissue first appears in the form of a band-like thickening of the epidermis called the mammary line or ridge. This line extends from the forelimb to the region of the hind limb in the embryo at 7 weeks. Most of the mammary line disappears soon after its formation except for a small portion in the thoracic region which persists and thickens to form the mammary primordium. The ectoderm of the primordium penetrates to the underlying mesenchyme and gives rise to between 16 and 24 small solid outbuddings. These canalize and form the lactiferous ducts and the alveoli of the mammary gland by the end of prenatal life. The lactiferous ducts open into an epithelial pit, which gives rise to the nipple as a consequence of proliferation of the surrounding mesoderm at term or later (Langman 1969). While the mammary ridge usually persists only in the midthoracic region in the human,

Table 1. Reported cases of ectopic breast of vulva

Case No.	Authors	Site/size	Age	Remarks
1.	Hartung (1872)	Labium majus, Lt/goose egg	30	Lactating change
2.	Woodruff & Seeds (1962)	Labium, Lt/–	39	Lactating change
3.	De Blasio (1905)	Labium majus, both/hen egg	young	Developed at puberty and enlarged during pregnancy
4.	McFarland (1930)	Labium, Rt/5cm	34	Discovered in 3rd pregnancy
5.	Mengert (1935)	Labium, Rt/2cm, Lt/1cm	24	Disovered in 5th pregnancy
6.	Levin & Diener (1979)	Globular labia majora	29	Primigravida
7.	Garcia et al (1978)	Labium majus, Rt/3cm 1cm, Lt/1cm 1cm	30	Primigravida lactating change
8.	Kaufman (1980)	Periclitoral/3cm	29	Mimicking periclitoral abscess, lactating change
9.	Bell (1926)	Labium majus, Lt/3cm	59	Discovered in 3rd pregnancy and later development of squamous carcinoma of pelvis
10.	Friedel (1932)	Unilateral	–	Fibroadenoma
11.	Fisher (1947)	Perineum, Rt/3cm	47	Fibroadenoma
12.	Siegler & Gordon (1951)	Labium majus, Lt/4cm	33	Fibroadenoma
13.	Burger & Marcuse (1954)	Vulva, Lt/2cm	48	Fibroadenoma
14.	Burger & Marcuse (1954)	Labium majus, Lt/2cm	30	Fibroadenoma with fibroadenomas of bilateral breast
15.	Two & Shanmugaratnan (1962)	Unilateral	–	Fibroadenoma
16.	Foushee et al (1967)	Labium, Lt/2cm	42	Fibroadenoma
17.	Foushee et al (1967)	Unilateral/3.5cm	24	Fibroadenoma
18.	Rickert (1980)	Labium majus, Rt/2cm	37	Intraductal papilloma, nullipara
19.	Greene (1936)	Labium, Rt/20cm	49	Adenocarcinoma with epidermoid carcinoma of vulva, nullipara
20.	Hendrix & Behrman (1956)	Labium Minus, Lt/3cm	58	Adenocarcinoma
21.	Guerry & Pratt-Thomas (1976)	Labium majus, Lt/1.5cm	62	Adenocarcinoma with infiltrating ductal carcinoma of bilateral breast
22.	Cho et al (1985)	Labium majus, Rt/4cm	70	Adenocarcinoma, metastasis to inguinal lymph node
23.	Fu & Reagan (1989)	–	–	Cystsarcoma phyllodes

nonresorbed remnants may give rise to an extra breast (polymastia) or nipple (polythelia) on occasion. Aberrant breast tissue is estimated to occur in 1 to 6% of the general population and appears to have a hereditary trait (Iwai 1907). Supernumerary nipples are relatively uncommon and they may also occur in males. Most cases are single, and about a third have more than two extra breast or nipples. Ectopic breast tissue is subject to physiologic and pathologic change just the same as in the normally situated breast. The breast shows morphologic changes of hormone response during menstrual cycle. The proliferative phase of the breast is characterized by small lobules with few terminal duct structures. Terminal duct epithelial mitoses are uncommon and the intralobular stroma is condensed and continues with interlobular stroma. The secretory phase of the breast shows an increasing size of the lobules, a number of terminal duct structures, duct epithelial basal vacuolization and mitosis. Intralobular stroma becomes increasingly loose and edematous. Perimenstural breasts undergo lobular contraction with the necorsis and sloughing of duct epithelium. there is a concomitant marked increase in the lobular stromal lymphocytic infiltrate and the metachromasia. Clinically, most of the lesions go undetected, as this tissue tends to remain quiescent until stimulated by hormone. They are occasionally recognized due to their enlargement or the discomfort during the latter part of the menstrual cycle (Garcia *et al.* 1978), and may be misdiagnosed as a periclitoral abscess (Reeves and kaufman 1980). More commonly, however, it is recognized as a consequence of the hormonal effect of pregnancy. The majority of patients are multiparous. The aberrant breast tissue was usually found above the umbilicus (Moore 1977). Cases below it, were extremely rare. Twenty-three cases of ectopic breast tissue of the vulva were reported and about one-third were bilateral (Table 1).

Ectopic breast tissue with or without lactating change was found in 9 cases, fibroadenoma in 8, cystosarcoma phyllodes in 1, intraductal papilloma in 1, and adenocarcinom in 4. The range of the vulvar mass size was from 1 to 7cm. The age of the patients varied from 24 to 70 years, and most of the malignant tumors occurred above fifty years of age.

Histologically, aberrant breast tissue consists of large ducts without fully developed lobules or terminal ductules. Stroma is often prominent, and may appear to be part of the lesion. Since the ectopic breast tissue is subject to the same physiologic and pathologic changes as the normally situated breast, the swelling and secretion of milk during pregnancy (Two and Shanmugaratnan 1962; O'Hara and Page 1985), and a variety of benign and malignant changes including fibrocystic diseases, fibroadenoma, intraductal papilloma (Ricket 1980), cystosarcoma phyllodes (Fu and Reagan, 1989) and carcinoma might occur. These pathologic changes found in aberrant tissue may be associated with those of normally situated breasts (Burger and Marcuse 1954; Guerry and Pratt-Thomas 1976).

Aberrant breast tissue has invariably been benign in the group of young women in whom the lesions were recognized during pregnancy or shortly thereafter. By contrast, 78% of those diagnosed during the non-gravid state were associated with neoplasia (Garcia *et al.* 1978). Fibroadenoma and adenocarcinoma tend to be more frequently seen in older patients.

In cases of the simultaneous presence of breast cancer, the possibility of metastasis from breast cancer was excluded by the presence of residual mammary tissue and intraductal carcinoma. Adenocarcinoma in aberrant breast tissue can metastasize to the regional lymph nodes (Cho *et al.* 1985).

During pregnancy, the optimum treatment may be that of simple observation or confirmatory biopsy, and such masses should probably be excised unless a significant regression takes place postpartum. Any vulvar mass in the older, non-pregnant woman requires prompt definite diagnosis and appropriate treatment.

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