

Case report:

Lactating Adenoma of Ectopic Breast Tissue in the Vulvar Region

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Abstract

Ectopic breast tissue is defined as glands located outside of the breast. Ectopic breast tissue has been thought to arise from a remnant of the embryonic mammary ridge along the milk line from the axilla to the groin, including the vulvar region. Vulvar tumors are uncommon and presence of aberrant breast tissue in this region is rare. Although lactation changes can occur in ectopic breast tissue, lactating adenoma in the vulva are extremely rare. Only few cases have been reported. We describe a case of a 24 year old woman who presented with vulvar mass first noted during her pregnancy. The mass was excised and histology revealed ectopic breast tissue and diagnosis of vulvar lactating adenoma was made.

Keywords: Milk line, Lactating adenoma

Background

Ectopic breast tissue may occur at any location along the milk line which extends from the axilla to the medial aspect of groin¹. Incomplete involution anywhere along the primitive milk streak can result in ectopic or accessory mammary tissue². Accessory breast tissue is most frequently localized in axillary region and only rarely observed at a vulvar site^{1,3}. Ectopic vulvar breast tissue displays the same pathologic and physiologic changes as normal breast tissue³. It can give rise to benign and malignant neoplasia but such cases are seen as a rarity². We present herein a case of unusual occurrence of ectopic breast tissue on the vulva presenting as a vulvar lactating adenoma in a pregnant female.

Case Description

A 24-year old primigravida was reported to our institution in April 2013 complaining of a vulvar mass of progressive growth. The patient was in 14th week of pregnancy. She had accidentally discovered the swelling when she was 12 weeks pregnant. The swelling was increasing in size gradually. The patient had no personal or family history of similar lesions. Initially a tentative diagnosis of lipoma was made on clinical examination. Fine needle aspiration cytology (FNAC) of the lesion was performed before coming to our institute and it was reported as suspicious of malignant lesion. Patient was referred to our institution. Physical examination revealed a tender, freely movable, firm 2x2 cm swelling in labium major of the vulva. No abnormalities of the

vulvar skin were noted and there was no inguinal lymphadenopathy. Uterus was compatible with 13 to 14 weeks' gestation, on clinical examination. Complete blood count and urinalysis were both normal.

The lesion was excised. Gross examination revealed a well circumscribed lobulated mass with a skin ellipse. It was measuring 2x2x1 cm. Cut surface of the lesion was tan with a thin fibrous pseudocapsule and no relation to overlying skin was seen. No necrotic or haemorrhagic foci were present. Histologic study showed a well circumscribed unencapsulated lesion composed of mammary glands with typical features of lactating adenoma (Fig. 1). Mammary glands were arranged in lobules separated by thin fibrovascular stroma. Glands were cystically dilated showing apocrine changes and secretory activity (Fig. 2). There was no evidence of atypia or mitoses. The diagnosis of a vulvar lactating adenoma was made.

Discussion

Ectopic or aberrant breast tissue is defined as mammary glandular parenchyma found beyond the anatomic extent of the breast⁴.

Ectopic breast tissue or accessory mammary glands arise when milk line remnants outside the pectoral region fail to regress¹.

Incidence of ectopic breast tissue is reported to occur in 2-6% of women². In humans, ectopic breast tissue occurs more frequently close to the breasts and a vulvar location is rare⁶. Ectopic breast tissue in the vulva was first identified by Hartung in 1872⁵. There are only a few case reports in the literature, of breast tissue arising in the vulva. This might be related to the fact that the vulva lies at the inferior end of the embryonic mammary ridge and therefore is a rare site for the location of ectopic mammary tissue². At present, controversy exists regarding the histological origin of vulvar lesions and the debate includes ectopic breast tissue,

cutaneous apocrine glands and most recently native mammary - like anogenital glands⁷. Mammary like tissue in vulva is currently considered to be ectopic in nature and distinction is made between ectopic breast tissue and mammary- like anogenital glands, which are located at a more medial site close to labia minora in greater numbers^{1,8}.

Ectopic breast tissue is subject to the hormonal influences of menstrual cycle, pregnancy and lactation⁵. Lesions become symptomatic during menarche, pregnancy and breast feeding¹. Woodruff and Seeds reported that more commonly aberrant breast tissue is recognized first during pregnancy manifesting as labial enlargement⁹. Ectopic or aberrant breast tissue displays same pathologic changes as in normally positioned breasts and there exists potential of developing benign or malignant neoplasia^{3,7}. Benign cystic diseases, fibroadenomas, lactating adenomas, intraductal papillomas, phyllodes tumor and adenocarcinoma of the mammary like tissue of the vulva have been described⁸.

Ectopic lactating adenoma has been reported and are seen as early as 23 and upto 39 years of age². Vulvar lactating adenoma is extremely rare and only few cases have been reported. Vulvar lactating adenoma may present with swelling or milk leakage through an injury². Pregnancy and lactation superimpose significant morphological change on adenomas of breast and ectopic breast tissue. Our patient was 24 years old primigravida presenting with a vulvar mass, increasing in size during 12th week of gestation with microscopic features of lactating adenoma. Histologically, ectopic lactating adenomas are indistinguishable from those of the breast. Vulvar lactating adenoma may be misdiagnosed as adenocarcinoma if breast tissue is not anticipated. For vulvar lesions, differential diagnosis includes vulvar carcinoma, Bartholin gland disorders, epidermal cyst and

crural hernia¹. Thus, differential diagnosis of vulvar mass in gravid women should include this entity as ectopic vulvar breast tissue, though rare, becomes symptomatic during pregnancy. A histopathologic study is must to rule out the presence of a neoplasm. Symptomatic breast tissue found in the vulva should be completely excised to prevent recurrence of symptoms and also there exists a risk of malignant conversion.

Vulvar ectopic breast tissue is rare. Neoplasms arising in this accessory mammary tissue are extremely rare and can present as a challenge for both clinician and pathologist. Diagnosis is confirmed by biopsy.

Fig 1: Mammary glands with lactational changes in lactating breast tissue (H&E, 400X)

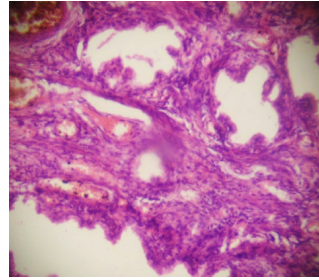
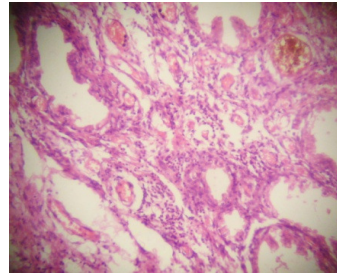


Fig 2: Cystically dilated glands with apocrine changes and secretory activity (H&E,400X)



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