Case Report

Fibroadenoma of the Vulva: A Case Report

Mohammad taghi Haghi ashtiani¹, Ali Zare mirzaie², Behrang Kazeminejad³

- 1. Dept. of Pathology, Tehran University of Medical Sciences, Tehran, Iran.
- 2. Dept. of Pathology, Iran University of Medical Sciences, Tehran, Iran.
- 3. Dept. of Pathology, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

ABSTRACT

Fibroadenoma of the vulva is an uncommon lesion. It has been suggested that they originate from vulvar mammary like other glands. A few examples of benign and malignant vulvar tumors arising from such glands have been reported.

A 32-year-old woman was referred with a subcutaneous mass at the vulva. The patient treated by simple excisional surgery with adequate peripheral margin.

Histological findings showed the characteristic features of fibroadenoma of vulva. Confirmatory IHC for GCDF 15 was performed which its result was positive. Up to our knowledge, only nineteen cases of vulvar fibroadenoma have been reported in English literature.

Key words: Vulva, Fibroadenoma

Introduction

Extramammary glands originated from embryonic milk line in the vulva can be the origin of several unusual tumors which are similar to breast lesions. This phenomenon is named as mammary-like anogenital gland (MLG) (1,2). These glands are considered to be natural structures of the vulva and it seems that they are closely related to eccrine glands (1,2). However, some investigations could not confirm that the theory of milk ridge which was proposed in the early years of 19th century can be applied to humans. It has been proved that primary mammary glands are not seen outside axillary regions and remain as natural structures of the vulva (1). Various but rare lesions may involve these glands. One of these lesions is vulvar fibroadenoma which its

incidence is not exactly known and few occurrences have been reported so far (3,4). This benign tumor presents histopathologic and IHC features similar to those of breast fibroadenoma (3). Reported cases include benign lesions treated by excisional surgery which is also used to treat breast fibroadenoma. However, few rare cases of phyllodes tumor have also been reported (3-5). An appropriate non-invasive technique which is applied for its diagnosis is fine needle aspiration (FNA) (5-7).

Case Report

A 32-year-old woman was referred with a soft tissue mass which was touchable and stemless and located in the subcutaneous area of the vulva. The mass which was attached to the skin of the vulva was

Received: 12 October 2007

Accepted: 3 December 2007

Address communications to: Dr Ali Zare mirzaie, Department of pathology, Iran University of Medical Sciences, Tehran, iran

Email: zaremir@gmail.com

about 2 cm in diameter. The lesion was painless, non-injured, and mobile and no sign of sinus, erythema or stretching was seen on the skin of the area. The mass was extracted and sent to the pathology laboratory.

Gross investigation showed a solid grayish cream mass 2.2 cm in diameter which was clearly bordered and showed a regular appearance on sectioning. Microscopic evaluation revealed epithelial and stromal proliferation in the fibromyxoid stroma. The epithelial structures composed of slit-like spaces (Figure 1) which were lined with bilayers of secretory epithelial as well as myoepithelial cells (Figure 2). No evidence of epithelial hyperplasia was observed. In some parts, papilla-like nodules were noticed, but none were real papillae and lacked a leaf-like appearance. The fibromyxoid stroma consisted of spindle cells with elongated nuclei, small nucleoli, and little cytoplasm. Its cellularity was rather low and no atypia or mitosis was seen. IHC study shows positive reactivity for GCDFF 15 in epithelial cells.



Figure 1. Slit-like glandular spaces in fibromyxoid stroma.(H&E X100)

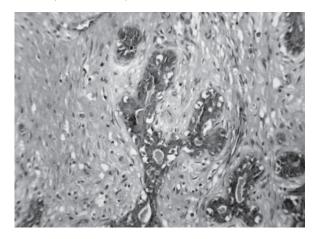


Figure 2. Bilayered lining of ductal structures composed of epithelial and myoepithelial cells.(H&E X400)

Discussion

Adnexal glands like mammary glands existing in the vulva and the skin of perianal region can be the origin of the lesions which are similar to breast lesions. Some of these benign lesions are as follows: Lactating gland fibroadenoma, papillary apocrine fibroadenoma, sclreosing adenosis, pseudoangiomatous stromal hyperplasia (PASH), mammary-like gland adenoma, and some kinds of carcinoma such as extramammary Paget's disease and invasive adenocarcinoma (1,7,8). These physiologic structures represent the characteristics of both apocrine and eccrine glands and create primary structures like those in tubulolobular units of the breast (1). As it was mentioned before, the theory of milk ridge is out of the question and Putte was the first one who criticized it (2). In his first report titled "mammary tissue of anogenital area", he discusses that the extension of mammary ridge to the vulva has not been proved definitively and the theory of milk ridge can not explain perineal tumors (2). In another experiment, Putte and Van Group suggested that specialized glands identical to mammary glands exist in the anogenital area (1). In fact, MLG is a natural part of vulvar structure and it seems that it bears a close relation to eccrine glands (1). As it was mentioned earlier, one of the benign lesions formed in these glands is fibroadenoma (3,4,9). Most of the reported fibroadenomas appear in one's thirties and forties (3,4,9). These rare benign tumors often manifest themselves as cutaneous or subcutaneous nodules with sharp borders (4). However, two of the reported cases presented with vulvar cysts (9). Bilateral tumors seldom occur and they might be accompanied by multiple malformations (10). Histologically speaking, they are characterized by stromal and epithelial proliferation (3,4,11). Most of the cases reveal an accompaniment of ectopic breast tissue in the form of ductular structures adjacent to the lesion or in direct histologic relation to the lesion. Therefore, it is suggested that they originate from ectopic breast tissue (3,6), but with regard to Putte's theory, the existence of normal breast tissue is not necessary for the foundation of these lesions (1,2).

The origin of these lesions is said to be the milklike glands of the vulva and we could not detect normal breast tissue in the lesion of our studied case either. Tumors which histologically resemble vulvar and perianal fibroadenomas which are found in these regions include perianal apocrine adenoma, adenoma of anogenital milk-like glands, and papillary apocrine

fibroadenoma (1,11,12). Phyllodes which is a tumor dependent on vulvar fibroadenoma is rarely seen in ectopic breasts. So far nine cases have been reported including vulvar, inguinal, and axillary tumors. They all presented with a painless, non-injured, and mobile lesion (5,13). Gross observation shows a mass with sharp borders and FNA reveals an appearance like fibroadenoma. However, histologic evaluation indicates a benign phyllodes tumor characterized by a proliferated stroma surrounding cystic spaces covered with flat or leaf-like epithelial cells. The epithelial coverage is a secretory type and dose not resemble mammary or anogenital glands. The stroma is hypercellular but scattered with mitosis. No malignant phyllodes with stromal overgrowth or necrosis have been reported yet (5,6,13). One of the important differential diagnoses of phyllodes tumor and vulvar fibroadenoma is mullerian adenosarcoma of cervix with an extension to the vulva. Other important differential diagnoses include adnexal lesions of the skin such as syringoma and papillary hidradenoma (6).

Another lesion which bears a close relation to fibroadenoma and phyllodes tumor is PASH (7). So far there cases of PASH in anogenital MLGs have been reported. All three cases were females aged between 40 and 55 years. Each case manifested itself through a single asymptomatic nodule which was 1.5-2 cm in diameter and located in the perineum, perianal region, and the labia major. The lesion in our study resembled breast PASH in appearance, i.e. it contained open slitlike canals which were anastomotic, empty of RBCs, covered with non-adjoining unclear cells lacking atypia and mitosis, and surrounded by a hyalinized collagenic stroma (7). A case of fibrocystic disease in anogenital MLG in a 25-year-old pregnant woman was reported which her pregnancy had worsened it (14). Initial diagnosis of fibroadenoma and other vuluar lesions is made by aspiration cytology which is considered as a non-invasive practical method to be carried out before the operation. However, the definite diagnosis to differentiate it from phyllodes is given after excisional biopsy (6).

IHC can also confirm the diagnosis. Other evaluated markers in the reported cases are as follows: ER, PR, SMA, S100, CK, and EMA (3,7). Since the IHC profile of the lesion was similar to breast fibroadenoma, GCDF-15, as it was assessed in our case, was also positive. This is 20th case of vulvar fibroadenoma which was reported.

References

- 1. van der Putte SC. Mammary-like glands of the vulva and their disorders. Int J Gynecol Pathol 1994 Apr;13(2):150-60.
- 2. van der Putte SC. Anogenital "sweat" glands. Histology and pathology of a gland that may mimic mammary glands. Am J Dermatopathol 1991 Dec;13(6):557-67.
- 3. Guler G, Usubutun A, Kucukali T. Fibroadenoma of the vulva. Arch Gynecol Obstet 2000 Apr;263(4):191-2.
- 4. Boscaino A, Sapere P, De Rosa GD. Fibroadenoma of the vulva. Report of a case. Pathologica 1996 Oct;88(5):444-
- 5. Oshida K, Miyauchi M, Yamamoto N, Takeuchi T, Suzuki M, Nagashima T, et al. Phyllodes tumor arising in ectopic breast tissue of the axilla. Breast Cancer 2003;10(1):82-4.
- 6. Mariappan MR, Lagera JE, Fadare O, Sibley RK. A 69-year-old woman with a vulvar lesion. Phyllodes tumor of the vulva. Arch Pathol Lab Med 2006 Jan;130(1):e11e12.
- 7. Kazakov DV, Bisceglia M, Mukensnabl P, Michal M. Pseudoangiomatous stromal hyperplasia in lesions involving anogenital mammary-like glands. Am J Surg Pathol 2005 Sep;29(9):1243-6.
- 8. Kinkor Z, Michal M. [Anogenital mammary type sclerosing adenosis--two case reports and the review of literature]. Ceska Gynekol 2004 Jul;69(4):292-7.
- 9. Baisre A, Heller DS, Lee J, Zheng P. Fibroadenoma of the vulva. A report of two cases. J Reprod Med 2002 Nov;47(11):949-51.
- 10. Degrell I. [Fibroadenoma in an accessory breast. A case of polythelia and fibroadenoma in the left breast region and a perivulvar accessory breast]. Fortschr Med 1979 Aug 2;97(29):1269-70.
- 11. Donati P, Amantea A. Adenoma of anogenital mammary-like glands. Am J Dermatopathol 1996 Feb;18(1):73-6.
- 12. Higgins CM, Strutton GM. Papillary apocrine fibroadenoma of the vulva. J Cutan Pathol 1997 Apr;24(4):256-60.
- 13. Chulia MT, Paya A, Niveiro M, Ceballos S, Aranda FI. Phyllodes tumor in ectopic breast tissue of the vulva. Int J Surg Pathol 2001 Jan;9(1):81-3.
- 14. Baykal C, Tulunay G, Usubutun A, Kucukali T, Ozer S, Demir OF. Fibrocystic disease of vulvar ectopic breast tissue. Case report and review of the literature. Gynecol Obstet Invest 2004;58(3):151-4.