Phyllodes tumor of the vulva – a rare diagnosis
Tumor filóide da vulva, um diagnóstico raro

Sara Proença*, Rita Lermann**, Amália Martins***, Carlos Veríssimo****
Hospital Beatriz Ângelo

Abstract

Mammary-like neoplasms have been described through the mammary-line, axilla, vulva and prostate, and are rare lesions of un uncertain histological origin. Phyllodes tumor occuring in the vulva is rare with only a few cases reported. We describe a case of a 41-year-old woman with a vulvar swelling that revealed to be a phyllodes tumor after complete excision.

Keywords: Phyllodes tumor; Vulva; Ectopic breast.

INTRODUCTION

Mammary-type fibroepithelial neoplasms of the vulva are rare lesions of uncertain histogenesis. Origins from ectopic breast tissue and from anogenital glandular tissue that shares similar histologic homology with breast tissue have been postulated[1].

Mammary-like tissue of the vulva can present as a small nodular swelling and a variety of breast diseases may arise. Lactating adenoma, fibroadenoma, pseudoangiomatous stromal hyperplasia and mammary type carcinomas have been described in the vulva[2]. Phyllodes tumor is a fibroepithelial neoplasms that account for less than 1% of all breast tumors and its occurence in the vulva is extremely rare[3].

We present a case of benign phyllodes tumor of the vulva in a 41-year-old woman. To our knowledge, this has only been described in literature in forteen other case reports[3-16].

Consent with patient was obtained in order to publish this case report.

*Interna de Ginecologia e Obstetrícia, Hospital de Cascais, Dr. José de Almeida
**Assistente Hospitalar de Ginecologia e Obstetricia, Hospital Beatriz Ângelo
***Assistente Hospitalar Graduada de Ginecologia e Obstetricia, Hospital Beatriz Ângelo
****Assistente Hospitalar Graduado de Ginecologia e Obstetricia, Diretor de Serviço de Ginecologia e Obstetrícia, Hospital Beatriz Ângelo

CASE REPORT

A 41-year-old woman was refered to evaluation for a vulvar mass. She complained of a vulvar tumor of progressive growth for the previous 9 months, not painful. She denied any acute pain, drainage or purulent discharge. Previous medical history was irrelevant. Physical examination revealed a well-circunscribed, 4x3cm, nodule located on the left labium majus. The tumor was firm and movable and not adhered to skin or other structures. Aspiration with needle was tried without success. The remainder of the gynecological and inguinal examination was normal. A simple excision was performed and during surgery the tumor was found to be a solid, firm mass, not adhered to other structures, well circunscribed and easily excised. Gross examination of the tissue revealed smooth-surfaced, pinkish-gray mass measuring 3,2 x 2,3 x 0,8cm with cystic projections and hypercellular stroma. Pathological examination showed a complete excision of phyllodes tumor on ectopic breast tissue. No recurrence has been detected after 6 months of follow-up.

DISCUSSION

Histological origin of mammary-type vulvar lesions remains uncertain. There is some controversy of its ectopic mammary tissue derived lesions, cutaneous apocrine glands and, most recently, lesions derived from mammary-like anogenital glands[1,17-19]. Ectopic breast

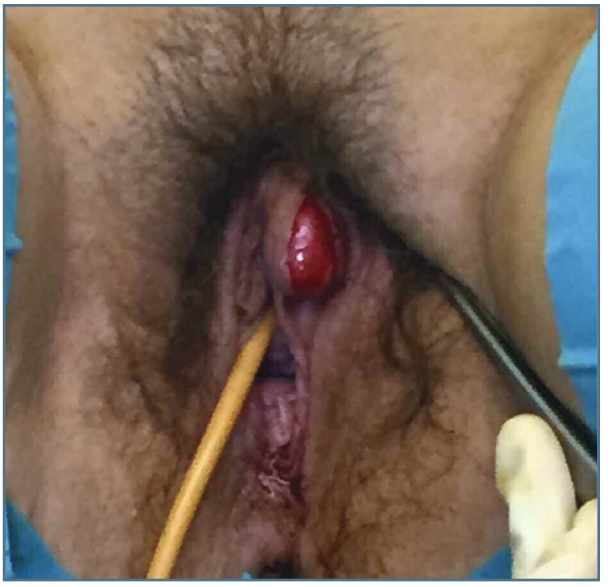

FIGURE 1. Tumor in the left labium majus

FIGURE 2. Macroscopic picture of the tumor

tissue has been the most frequently cited source, but adjacent extralesional ectopic breast tissue has only been poor documented[1]. Nonetheless, both ectopic breast tissue and mammary-like anogenital gland are susceptible to physiologic, dysplastic and malignant changes and its clinical presentation and subsequent behavior is comparable to lesions derived from the breast. There have been reported cases of benign and malignant lesions in the vulva such as invasive ductal carcinoma, ductal carcinoma in situ, lobular carcinoma, sarcoma, inflammatory carcinoma, adenocarcinoma, phyllodes tumor and fibroadenoma[5,19].

Phyllodes tumor of the vulva is histologically similar to phyllodes tumor of the breast[6]. There are some reports of its occurence in the axilla and prostate[7]. It is classified in benign, borderline or malignant according to its cellularity and mitotic activity.[11] Most patients present with a painless cystic vulvar swelling or nodule[4]. Most tumors are located in labium majus but other locations described include interlabial sulcus, mons pubis and periclitorial sites[2]. Reports range from solitary tumors to bilateral and 2 separate nodules. Current mainstray of therapy is surgical ressection with clear margins, being definitive in most cases[17].

Due to its rarity there is little information about the natural course of this disorder and prognosis is uncertain. Local recurrence seems to be the most common complication and there are no reports on metastatic tumors[5,10,13]. Since accurate diagnosis previous to the excision is rare, a proper surgical margin is needed to avoid local recurrence, or if needed, a re-excision for proper margins[10,16].

Our case was diagnosed as a benign phyllodes tumor after surgical excision, nonetheless, free margins were obtained and local recurrence was not documented when last seen, at 6 months of follow-up. As the prognosis is unclear because of the rarity of the tumor and paucity of published data on this rare tumor, close follow-up is warranted, irrespective of histologic categorization, and patient will be avaluated further on.

REFERENCES

1. Carter JE, Mizelli KN, Tucker JA. Mammary-type fibroepithelial neoplasms of the vulva: a case report review of the literature. J Cutan Pathol 2008:35;246-249.

2. Lee S, Nodit L. Phyllodes Tumor of Vulva. Arch Pathol Lab Med 2014:138;1546-1550.

3. Ozbudak IH, Akkaya H, Akkaya B, Erdogan G, Pestereli HE, Karaveli FS. Phyllodes Tumor of the Vulva: Report of 2 Cases. Turk Patoloji Derg 2013:29;73-76.

4. Salmo E, Shaw G, Tagore V. Phyllodes Tumour of the Vulva: Report of Case and Review of Literature. The Internet Journal of Pathology 2004:4 Number2.

5. Tbakhi A, Cowan DF, Kumar D, Kyle D. Recurring Phyllodes Tumor in Aberrant Breast Tissue of the Vulva. Am J Surg Pathol 1993:17;946-950.

6. Chulia MT, Paya A, Niveiro M, Ceballos S, Aranda FI. Phyllodes Tumor in Ectopic Breast Tissue of the Vulva. Int J Surg Pathol 2001:9;81-83.

7. Oshida K, Miyauchi M, Yamamoto N, Suzuki M, Nagashima

T et al. Phyllodes tumor arising in ectopic breast tissue of the axilla. Breast Cancer 2003:10;82-84.

8. Mannan AA, Kahvic M, Aziz AH. Phyllodes tumor of the vulva: Report of a rare case and review of the literature. Am J Dermatopathol. 2010 Jun;32(4):384-386.

9. Tressera F, Grases PJ, Izquierdo M, et al. Fibroadenoma phyllodes arising in vulvar supernumerary breast tissue: report of two cases. Int J Gynecol Pathol. 1998;17:171-173.

10. Hefferman TP, Sarode VR, Hoffman B, Lea J. Recurrent phyllodes tumor of the vulva: a case report with review of diagnostic criteria and differential diagnosis. Int J Gynecol Pathol. 2010 May;29(3):294-297.

11. Giger OT, Lacoste E, Honegger C, Padberg B, Moch H, Varga Z. Expression of the breast differentiation antigen NY-BR-1 in a phyllodes tumor of the vulva. Virchows Arch 2007 Apr;450(4):471-4. Epub 2007 Feb 21.

12. Mariappan MR, Lagera JE, Fadare O, et al. A 69-year-old woman with a vulvar lesion. Arch Pathol Lab Med. 2006;130:e11–e12.

13. Kazakov DV, Spagnolo DV, Stewart CJ, et al. Fibroadenoma and phyllodes tumors of anogenital mammary-like glands: a series of 13 neoplasms in 12 cases, including mammary-type juvenile fibroadenoma, fibroadenoma with lactation changes, and neurofibromatosis-associated pseudoangiomatous stromal hyperplasia with multinucleated giant cells. Am J Surg Pathol. 2010;34(1): 95–103.

14. Fu L, Lau S, Roy I, Ferenczy A. Phyllodes tumor with malignant stromal morphology of the vulva: a case report and review of the literature. Int J Gynecol Pathol. 2011;30(2):198–202.

15 - Kais M, Bitterman-Deutsch O, Schwartz J, et al. Phyllodes tumor in aberrant breast tissue of the vulva. Cervix 13:105--106,1995.

16. Denlinger LN, Lokhandwala PM, Abendroth CS. Benign Phyllodes Tumor of the Vulva: A Case Report and Literature Review. Rare Tumors. 2015;7(4):6010. doi:10.4081/rt.2015.6010.

17. Cantú de Leon D, Montiel DP, Vázquez H, Hernández C. Cetina L, Lucio MH. Vulvar Fibroadenoma: a common neoplasm in an uncommon site. World J Surg Oncol 2009:7;70-73.

18. Putte SC. Mammary-like glands of the vulva and their disorders. Int J Gynecol Pathol. 1994:13;150-160.

19. Butler B, Leath III AA, Barnett JC. Primary invasive breast carcinoma arising in mammary-like glands of the vulva managed with excision and sentinel node biopsy. Gynecol Oncol Reports 2014:7;7-9.

ENDEREÇO PARA CORRESPONDÊNCIA
Sara Proença
Hospital de Cascais Dr José de Almeida
Lisbon, Portugal
E-Mail: saritaproenca@gmail.com

RECEBIDO EM: 14/03/2017
ACEITE PARA PUBLICAÇÃO: 10/05/2017