

Recurrent vulvar breast fibroadenoma: presentation of a rare clinical condition

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Abstract

Vulvar fibroadenoma is an ectopic mammary-like fibroepithelial tumor of undetermined pathogenesis. Surgery is usually the primary option of treatment for this condition. Recurrent vulvar fibroadenoma is a rare clinical condition, and it has therapeutic challenges for surgeons and pathologists. Three cases of recurrent vulvar fibroadenoma were reviewed and studied (at 25, 39, and 18 years old for each case). We summarize and report our clinical experience on recurrent vulvar fibroadenoma. All patients were surgically managed with a satisfactory outcome and were followed up for at least 6 months. This is the first case series to address a rare clinical condition in vulvar fibroadenoma. Tumor size, conditions of high hormonal exposure, iatrogenic issues, and residual lesions contribute to recurrence. The standard surgical strategy for vulvar fibroadenoma is important for lowering the risk of local recurrence.

Keywords

Fibroadenoma, vulva, breast neoplasms, recurrence, tumor, lesion

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Introduction

Ectopic breast tissue, a residual tissue in embryological development, occurs in approximately 5% of the female population.¹ This tissue appears along the milk line, frequently near the normal mammary gland, but rarely occurs inferiorly below the

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inguinal region.² Therefore, the vulva is an uncommon location for ectopic mammary tissue. Vulvar fibroadenoma is an ectopic mammary-like fibroepithelial tumor, for which surgery is usually the paramount therapeutic consideration. Recurrent vulvar fibroadenoma is a rare clinical condition, which can be multifactorial. In this case series, we summarize our experience of recurrent vulvar fibroadenoma in several patients who were surgically treated and who showed local recurrence.

Clinical presentation

Case one

A 25-year-old young woman was admitted to hospital in December 2017 complaining of left vulvar recurrent swelling for 10 months. The mass was painless, but progressively growing in size. She had a previous history of vulvar breast fibroadenoma, which was surgically removed 4 years ago. She was free from recurrence until a swelling of the same location was discovered during pregnancy.

A physical examination showed a $5 \times 2 \times 2$ -cm mass located beneath the scar at the left lip of the labium majus. The mass was soft, movable, and well-defined. No abnormalities were found by abdominal and gynecological examinations. Neither fine-needle aspiration cytology nor computed tomography was used because ultrasonography indicated a benign lesion. At this stage, tumor recurrence was noted and we decided to proceed with broad tumor excision. The operation went smoothly under local anesthesia. Frozen sections of tumor tissue were cut and showed vulvar fibroadenoma. The incision was routinely closed. The patient was discharged the next day after surgery. The patient was free of detectable lesions at a 6-month follow-up.

Case two

A 38-year-old woman complained of vulvar swelling for 1 month and was admitted to hospital in November, 2010. The patient was also scheduled for *in vitro* fertilization cycles for infertility. A physical examination showed a $3 \times 1.5 \times 2$ -cm, soft, and movable mass located in the mons pubis. Ultrasound and fine-needle aspiration suggested the diagnosis of ectopic breast fibroadenoma. We successfully performed surgical resection 1 week after which the patient underwent ovarian stimulation. The patient failed to become pregnant. She started a new stimulation 3 months after the first attempt. However, she experienced failure again. She underwent an ultrasound follow-up, which showed that a $1.2 \times 1.0 \times 1.0$ -cm nodule had recurred in the same location. Because of the limited size of recurrence, conservative observation was recommended. She started the third *in vitro* fertilization attempt, and became pregnant this time. At 20 weeks of gestation, the mass became apparent, palpable, and was growing aggressively in size. At 38 weeks of gestation, she underwent cesarean section, followed by tumor resection at the same stage. A gross examination indicated a tumor that measured $5.5 \times 4.5 \times 4$ cm, with a pale, white, lobulated cut surface. Pathology showed histological features in accordance with those of breast fibroadenoma. The patient has been followed up annually. Ultrasonography reported a steady lesion of 6 mm in diameter, which was attributed to a benign disease. She is currently under conservative observation.

Case three

A 16-year-old girl complained of a vulvar mass for several months and was admitted to hospital in June, 2008. On a physical examination, a soft and movable nodule was palpable in the left labium majus.

Perineal ultrasonography showed a well-defined, hypoechoic mass that measured $2.1 \times 1.5 \times 1.5$ cm in size. Because of psychological anxiety, the tumor was resected, with a pathological diagnosis of ectopic breast fibroadenoma.

Four months later, she discovered a palpable mass of $2.5 \times 2.5 \times 2.0$ cm in size beneath the scar and the lump was removed again. One and a half years later, vulvar swelling of $3.7 \times 3.2 \times 2.5$ cm was found for the third time, and the girl underwent broader resection with a safety surgical margin of 1.5 cm. The patient has been under regular follow-up for 8 years without recurrence.

Ethics statement

Approvals from the Ethical Committees of Guangdong General Hospital, Guangzhou Women and Children's Medical Center, and Guangdong Provincial Hospital of Chinese Medicine were obtained before initiating this study. Written informed consents for the use of clinical data, images, and permission for publication were obtained from the patients.

Ultrasound

Ultrasound features of ectopic breast fibroadenoma are specific. A hypoechoic solid mass can be found subcutaneously,

with typically a clear border, well-defined margin, membranous echo, and posterior echo enhancement. Sometimes the internal echo is not homogenous, with sporadic hyperechoic spots. Nutrient vessels of the mass are hypoechoic and are distributed in a reticular manner (Figure 1). Notably, a higher incidence of malformations in the urinary tract, such as polycystic kidneys and ureteric obstruction, can be seen in those with ectopic breast tissue.³ Urinary ultrasonography was performed in our cases, but no congenital anomalies in the urinary tract were found.

Pathology

On gross examination, typical ectopic breast fibroadenoma is an encapsulated, solid, gray-white, lobulated tissue, with a smooth surface. Epithelial and mesenchymal proliferation can be seen. The proportion of glands and stroma is relatively consistent through the mass. Most of the growth pattern is in the pericanalicular style, in which the stroma surrounds glands with an open lumina. The glands have dual layers, including an inner epithelial and outer myoepithelial layer. The epithelium is composed of a single layer with cuboidal-to-columnar cells. Sometime the stroma shows myxoid changes without calcifications (Figure 2). In our cases,

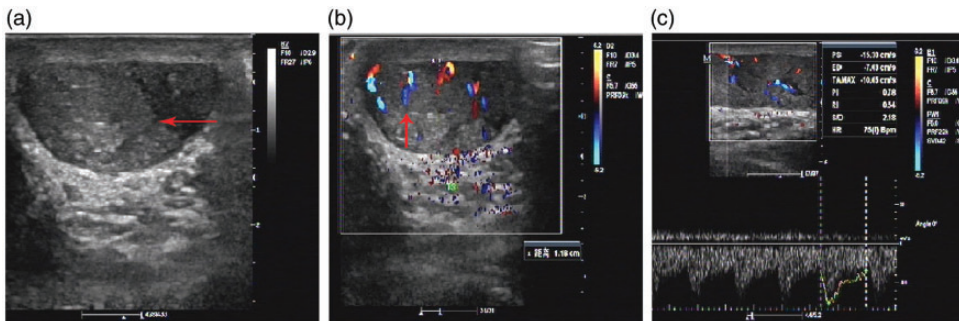


Figure 1. Sonographic images show typical features of vulvar fibroadenoma. (a) Hypoechoic mass with a clear border and well-defined margin (red arrow); (b) hypoechoic nutrient vessels in a reticular manner (arrows); and (c) measurement of the resistance index of nutrient vessels in the mass.

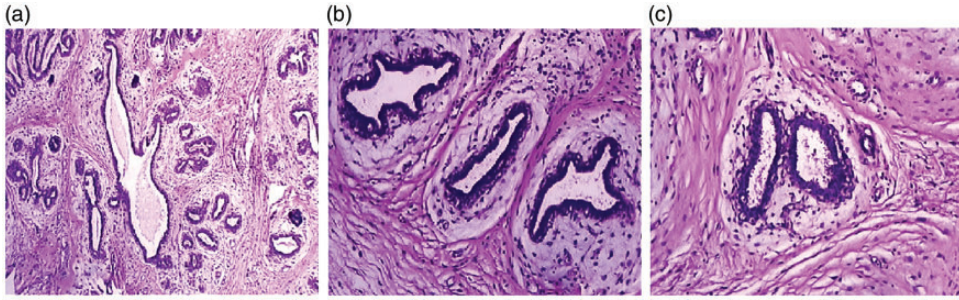


Figure 2. Microphotograph shows typical histological features of vulvar fibroadenoma. (a) Hematoxylin–eosin staining ($\times 40$) shows epithelial and mesenchymal proliferation of which the growth pattern is the pericanalicular style; (b) glands have dual layers composed of an inner epithelial and outer myoepithelial layer; and (c) the epithelium is composed of a single layer with cuboidal-to-columnar cells.

immunohistochemical studies were not necessary after confirmation of benign lesions.

Discussion

A description of ectopic breast tissue in the vulva was first described in 1872.⁴ However, only a limited number of such cases have been reported in the literature.⁵ We reviewed the literature in MEDLINE (via Pubmed), EMBASE, and LILACS, by using “(recurrence [Title/Abstract]) AND (vulvar fibroadenoma [Title/Abstract]) OR ectopic breast fibroadenoma [Title/Abstract]) OR ectopic breast tissue [Title/Abstract]) OR aberrant breast tissue [Title/Abstract])” to create the search. We found no related papers. This is the first series to address this rare clinical condition.

Multiple factors might contribute to recurrence of vulvar breast fibroadenoma. Based on our cases, tumors > 2 cm are associated with a higher risk of recurrence. Latent vulvar breast tissue might then be triggered to proliferate during hormonal stimulating periods, such as pregnancy, lactation, or puberty. Furthermore, a residual lesion is likely relevant to recurrence. There is no rule for mastectomy or quadrantectomy because the particular location of the vulva limits the extent of excision. Therefore, a definite resection with a clear

and safe margin is crucial to avoid local recurrence. Finally, some iatrogenic factors, such as ovarian stimulation therapy, might lead to tumor recurrence.

The recurrence rate of vulvar breast fibroadenoma after excision is difficult to establish because of its rarity. The recurrence rate is reported to be lower than 3% in surgically treated orthotopic fibroadenoma.⁶ Famá et al.⁷ retrospectively reported a large series of 327 patients with ectopic breast resections, and only one recurrence was observed. The recurrence rate of vulvar breast fibroadenoma might be slightly higher than that of orthotopic lesions because incomplete removal occurs more easily.

Not only benign lesions, such as cysts, intraductal papilloma, and phyllodes, but also malignancies, such as invasive carcinomas and extra-mammary Paget’s diseases, should be taken into consideration for differential diagnosis.^{8,9} Because this disease develops beyond the normal breast, the diagnosis may be postponed without a high index of alertness. The prognosis of ectopic breast carcinoma appears to be poorer than malignancy that develops in normal breast tissue because involvement of lymph nodes might be earlier.⁷

Management of vulvar breast fibroadenoma ranges from simple observation to

broad resection. Small-sized lesions or menopausal lesions may regress spontaneously. Surgery is indicated for lesions that are > 2.5 cm in diameter, fast-growing, painful, and those distorting vulvar architecture. Those with stromal hypercellularity and those with malignant suspicions are also indications for a definite surgical excision. For patients who are exposed to risk factors of disease development, more aggressive treatment with close follow-up is warranted.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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