International Journal of Reproduction, Contraception, Obstetrics and Gynecology
Ilayappan SS et al. Int J Reprod Contracept Obstet Gynecol. 2022 Jul;11(7):2046-2048
www.ijrcog.org
pISSN 2320-1770 | eISSN 2320-1789

DOI: https://dx.doi.org/10.18203/2320-1770.ijrcog20221695

Case Report

A rare case report of vulval fibroadenoma

Saranya Sri Ilayappan[1], Sujitha Sivarajan[1]*, Mala Vijayakrishnan[1], Sarah Grace Priyadharshini[2]

[1]Department of Obstetrics and Gynaecology, [2]Department of Pathology, Vijaya Hospital, Chennai, Tamil Nadu, India

Received: 17 May 2022
Accepted: 06 June 2022

***Correspondence:**
Dr. Sujitha Sivarajan,
E-mail: drsujitha23@gmail.com

ABSTRACT

Vulval fibroadenoma is an extremely rare entity seen in women of the age group 20-80 years. Only 50 cases of ectopic benign mammary tissue have been reported in medical literature, with vulva being the second most common extra-mammary site. We report a case of a 44 years old female, para 2 live 2, who presented with an initially endophytic swelling in her vulva for 6-7 months, which became exophytic since 2 months, associated with pain for 1 month and purulent discharge since 2 days. On examination, a 4×2 cm fleshy exophytic mass with a cauliflower-like appearance was noted on the right labia majora, just below labia minora with no active purulent discharge. Based on the clinical examination, our differential diagnosis included condyloma accuminata, infected sebaceous cyst, hidradenoma papilliferum, apocrine adenoma, Bartholin's cyst and phyllodes tumour. A surface biopsy and cytology taken from the growth was suggestive of infected Bartholin's cyst. Pus culture showed no growth. She underwent excisional biopsy subsequently with 2 mm margin clearance. Histopathology was reported as vulval fibroadenoma with stromal cellularity. A unique feature of our case is that an initially endophytic mass became exophytic with purulent discharge unlike many other reported cases worldwide. A surface biopsy is a useful tool to arrive at a preoperative diagnosis. Surgical excision with a clear and safe margin is recommended to avoid recurrence. If malignancy is reported in histopathology, testing for hormone receptors can be helpful. Women have to be generally more aware about such abnormal lesions and should report immediately.

Keywords: Vulval fibroadenoma, Ectopic mammary tissue, Condyloma accuminata

INTRODUCTION

The vulva is an ill-defined area in females comprising of mons pubis, clitoris, labia majora, labia minora, urethral orifice, anal orifice and perineal body. The labia majora contains sebaceous glands, sweat glands and hair follicles which keeps it soft and moist. The presence of these structures in labia majora renders them liable to common skin lesions like sebaceous cyst, folliculitis and furunculosis. Notably, Bartholin's gland lies postero-laterally to the vaginal orifice, deep to the bulbospongiosus muscle and superficial to the outer layer of the triangular ligament. It is a compound racemose gland, that secretes lubricating mucus during coitus. It is normally not palpable when healthy but can be readily palpated between the finger and the thumb when inflamed. Keeping in mind the vulval anatomy, the lesions or tumours found in the vulva can also have an ectopic origin as in ectopic mammary tissue. Mammary glands are bilateral modified sweat glands each containing 15-20 lobes. Each lobe is made up of acini, ducts and fat with the ducts opening into the nipple. Ectopic mammary tissue has been reported worldwide over the last 50 years, with vulva being the second most common extra-mammary site and axilla being the first common site. It is seen in women of age group 20-80 years.[4] Most common theories given for this condition are milk line theory and mammary like anogenital glands (MLAG) theory. Medical literature shows very limited case reports on this condition.

CASE REPORT

A 44 years old woman, para 2 live 2 presented to our outpatient gynaecology department at Vijaya hospital with complaints of vulval swelling for 7-8 months, along with purulent discharge from the mass for 2 days prior to outpatient consultation with us. We elicited history of an endophytic growth in the right labia majora for an initial period of 4-5 months. This was followed by a break in the epithelial layer on the most prominent point of the swelling, through which the growth became exophytic, associated with pain. There was no itching. On examination, a 4×2 cm exophytic mass with a cauliflower like appearance was noted on the right labia majora, just below labia minora, which was firm in consistency, not tender and not attached to underlying structures, as can be seen in Figure 1. No active purulent discharge was noted. Abdominal and pelvic examination was found to be normal. Pap smear was negative for intraepithelial or invasive malignancy. We had a differential diagnosis of condyloma accuminata, hidradenoma papilliferum, phyllodes tumor, Bartholin's cyst and sebaceous cyst. Hence, surface biopsy and cytology were taken. It was suggestive of infected Bartholin's cyst and cytology shows no abnormality. Pus culture showed no growth. She underwent excision of the mass with 2 mm margin clearance. Cut section showed no necrosis or suspicious area. The histopathology report was reported as fibroadenoma with stromal cellularity arising from mammary-like glands of the vulva. Microscopy showed stratified squamous epithelium overlying a neoplasm composed of tubular and elongated ducts lined by bilayer epithelium in a cellular fibrous stroma with spindle cells, as shown in Figure 2. There was no stromal mitosis. Deep margin was free of tumour. Post operative period was uneventful. She was followed up and counselled.

Figure 1: Gross appearance of an exophytic cauliflower like growth from right labia majora just below labia minora.

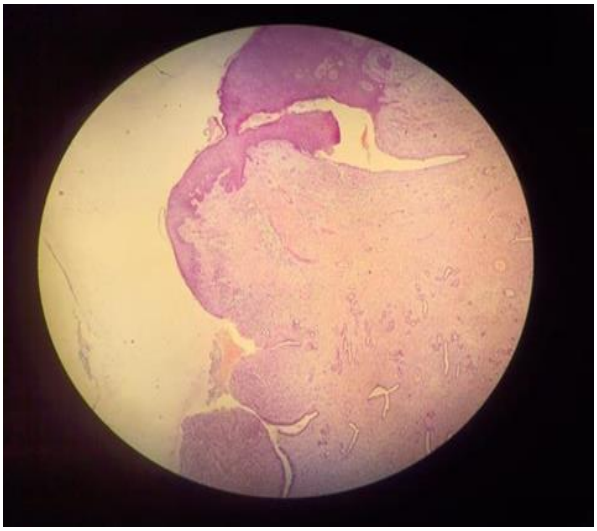

Figure 2: Fibroadenoma with stromal cellularity arising from mammary like glands of vulva, Section shows stratified squamous epithelium overlying a neoplasm composed of tubular and elongated ducts lined by bilayered epithelium in a cellular fibrous stroma with spindle cells.

DISCUSSION

The first ever case of ectopic mammary gland tissue was reported by Hartung in 1872. Since then there have been very few reported case studies of ectopic mammary tissue. Accessory breast tissue is seen in 5% of Japanese women. This has several forms and they are classified by Kajava as: 1) Complete breast with nipple, areola and glandular tissue; 2) supernumerary breast without areola but with nipple and glandular tissue; 3) supernumerary breast without nipple but with areola and glandular tissue; 4) aberrant glandular tissue without nipple or areola; 5) pseudo-mammary with nipple and areola but without glandular tissue, 6) polythelia, presence of nipple only; 7) polythelia areolaris, presence of areola only; 8) Polythelia pilosa, presence of patch of hair only. While aberrant breast tissues are seen along the milk line, other rare presentations include vulva, shoulders, buttock and ears.

The histogenesis of vulval fibroadenoma is a much-disputed topic. There are two proposed theories; one theory is based on fetal embryogenesis. According to this theory when the embryo has a crown-rump length of 7 mm i.e., at gestational age of 5 weeks, milk line appears, extending from the axilla to groin in a curvilinear pattern. Over time the milk line involutes to form an epidermal thickening in the thoracic region which develops into the breast. In case of incomplete involution ectopic mammary tissue can be found anywhere along the milk line. Vulval ectopic mammary tissue would be seen at the inferior end. The most common problems encountered in an ectopic breast tissue include fibrocystic changes, mastitis, fibroadenoma, and carcinoma. The second theory was postulated by Putte in 1994. He suggested the presence of mammary like glands in the anogenital area which is

closely related to eccrine glands. These glands located in the vulva are mammary like anogenital glands (MLAG). Similarly in our case fibroadenoma with stromal cellularity has been noted to arise from mammary like glands of vulva. The possibility of benign or malignant nature of the lesion or lactational changes of the ectopic breast tissue or MLAG are attributable to the presence of hormone receptors. These lesions are usually identified in dormant ectopic mammary gland tissue as a result of hormonal changes associated with puberty, pregnancy and lactation.

The most common presenting symptom in this condition, as reported by cases worldwide, is vulval swelling. In our case, apart from vulval swelling, the patient complained of purulent discharge, similar to a study by Ahamed et al.[3] While most of the cases reported an endophytic mass, in our case initially it was an endophytic mass which became exophytic later.

One has to consider differential diagnosis ranging from benign to malignant epidermal cyst, Bartholin's cyst, lipoma, phyllodes tumor, fibrocystic disease, apocrine adenoma, hidradenoma papilliferum, extra-mammary Paget's disease and mucinous adenocarcinoma. In our case, while the gross appearance of the mass gave us an impression of possible malignancy, the final histopathology report diagnosed it as benign vulval fibroadenoma.

Owing to the rarity of this condition, the recurrence rate of vulvar fibroadenoma after excision is difficult to establish.[4] Li et al has discussed 3 cases of recurrent vulvar fibroadenoma.[2] Tumour size more than 2 cm, conditions of high hormonal exposure like pregnancy, lactation or puberty, iatrogenic issues like ovarian stimulation and residual lesions are said to contribute to recurrence. Hence, it is crucial to ensure a resection with a clear and safe margin. This also aids in adequate histological review of surrounding tissue.

CONCLUSION

To conclude, vulval fibroadenoma is a rare entity. While there is no specific screening method to identify such lesions, women have to be general more aware about such abnormal lesions and report immediately. Surgical excision is the mainstay of treatment for vulval fibroadenoma. One has to consider testing for hormone receptors if malignancy is confirmed. This condition is also known to have a good prognosis. Follow-up care for the patient should also be explained in detail.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Ilayappan SS, Sivarajan S, Vijayakrishnan M, Priyadharshini SG. A rare case report of vulval fibroadenoma. Int J Reprod Contracept Obstet Gynecol 2022;11:2046-8.