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Case Report

Leiomyoma Arising from the Vulva: A Case Report and a Brief Clinico-Pathological Review of Benign Vulvar Tumors

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ABSTRACT

Leiomyoma is a benign smooth muscle tumour. Although rare in the vulva, the tumours are common in the uterus in women of reproductive age.

A 32-year-old woman presented with a painless, firm, well circumscribed vulvar mass of three years' duration. Based on its location, appearance, and being fairly common, Bartholin's gland cyst was readily considered as the tentative diagnosis before surgery. The histopathology of the tumour excised at surgery showed leiomyoma. Although the condition is rare, early biopsy of all solid and partially solid vulvar masses is highly recommended to confirm diagnosis and arrest progression of benign lesions into malignancies.

Keywords: Vulvar, Leiomyoma, Tumor

Introduction

The cells of the vulva have the inherent potential of proliferating into benign and malignant tumors. Leiomyoma of the vulva is a benign smooth muscle tumor. It is quite rare globally. African regional case report publications are scanty.[1,4,5] The risk factors for its development are not well articulated.[2] The common site of occurrence for leiomyoma is the uterus.[3] Affected women with vulvar tumors rarely seek medical treatment during early stages of growth because the condition is often painless.[2] This reluctance in seeking medical help may contribute to the apparent rarity of the condition. The authors hereby present this rare case of vulvar leiomyoma recently managed at our centre, with comments on differential diagnoses of common benign vulvar tumors.

Case Report

The woman, a 32-year-old nullipara, presented at the gynecology clinic with a three-year history of right vulvar swelling. The swelling had increased in size over the period but remained painless. There were no associated abdominal or gynecological symptoms. She had no relevant past medical, family, or social history.

She was clinically well, and her vital signs were stable. Her chest, cardiovascular and abdominal

examinations showed normal findings. Pelvic examination revealed a well circumscribed round to oval mass at the posterior of the right labium majus, half-way between the clitoris and the posterior commissure of the labia minora, measuring 6cm x 5cm x 4cm in dimensions (Figure 1). It was non-tender, partly cystic, and relatively mobile under the vulvar skin. A pelvic ultrasound scan was done, and the result was normal with no detectable pelvic pathology.

A diagnosis of Bartholin's cyst was made, and the patient was counselled for surgery and excision of Bartholin's cyst. Her full Blood Count was normal with a Hemoglobin of 13.3g/dL, and Hb Genotype AA. Retroviral (HIV) screen and the hepatitis B surface antigen (HBsAg) screen were negative.

Surgery was performed under spinal anaesthesia. After aseptic skin cleansing and draping in Lithotomy position, a Foley's catheter was inserted to drain the bladder during surgery. The vulvar skin was held firm over the mass with forceps. A longitudinal incision was made on the skin, lateral to the right lip of the labia minora, and developed to the plane of cleavage. The mass was shelled out whole, and the cavity obliterated with multiple vicryl sutures. Blood loss was minimal. The urethral catheter was removed after surgery. The excised tumor was sent for histopathologic diagnosis. The patient was discharged home the following day on post-operative antibiotics and analgesic medications.

She was seen and reviewed on her third post-operative day, and at weekly intervals for three weeks. The vulvar wound healed well, and she was seen monthly as an outpatient. After two visits she was counselled for prolonged three-monthly follow-up outpatient visits, in the next year.

- **Histopathology Report**: Grossly, it was received fixed in 10% buffered formalin. It was partially encapsulated, round to oval, pale grey and firm in consistency. (Figures 2[a] and 2[b]) It measured 5cm by 4cm by 3cm. Serial cut sections show uniformly solid grey-white surfaces with no cystic areas. (Figures 3[a] and 3[b]).

  Microscopy showed fascicles of regular spindle-shape smooth muscle cells cut in various planes with no cytologic atypia. (Figures 4a, 4b, 4c). Mitotic figures were few and far between. A diagnosis of vulvar leiomyoma was made.

Discussion

The vulva is the visible part of the female external genital organs and collectively, it includes the labia majora and labia minora, the clitoris, the vestibule, the bulb of the vestibule and the Bartholin's gland.

Clinically, the tentative diagnoses of a vulvar mass in a female of reproductive age should include Bartholin's gland cyst or duct cyst, Bartholin's gland abscess, and epidermal inclusion cysts. Other benign vulvar conditions such as hidradenoma, papilloma, fibroma, fibroepithelial polyp, and angioma, may present as painless round-to-oval benign vulvar tumors and should be considered in the differential diagnoses of a vulvar mass.

Vulvar leiomyomas are rare tumors.[1,2,4,5,] Very few case-report publications are on record from far-flung regions globally including reports from Sokoto, Nigeria,[3] Dallas, USA[4] and a recent comprehensive meta-analysis review article by Chao Sun et al, from Shaanxi, China.[2] Vulvar tumors are reported to occur in the fourth and fifth decades of life[5] and various histologic types have been described, which include spindled, epithelioid and myxoid neoplasms.[6] Cases have been reported in other rare "ectopic" sites such as the ovaries, the clitoris and the retroperitoneal areas.[7,8] They account for 0.03% of all cases of gynecological tumors,[9] and may arise from the dartos muscle of the labia majora, from the smooth muscle cells around vascular channels or from muscles of hair follicles.[10] Histologically, the differential diagnoses of vulvar leiomyoma include leiomyosarcoma, neurofibroma, dermatofibroma, and lipoleiomyoma.

Leiomyoma and leiomyosarcoma have similar immunohistochemistry. Both stain positively for desmin, vimentin, and smooth muscle actin (SMA). They can be distinguished by using the criteria

proposed by Nielson et al.[11]

According to Nielson et al, sarcomas are mostly more than 5cm in diameter, have up to and above 5 mitotic figures per 10 high power fields, exhibit cytologic atypia, and have unencapsulated, infiltrative borders. Neurofibromas differ from leiomyoma by being unencapsulated, having nerve sheath cells, fibroblasts, with the characteristic mast cells background, and showing areas of epidermal atrophy with indistinct rete ridges. Dermatofibroma shows proliferation of fibroblasts with thick mesh of collagen bundles, especially at the margins. Factor XIIIa stain is mostly positive for dermatofibroma. Lipoleiomyoma has a preponderance of adipocytes, in comparison with leiomyoma.

In addition to leiomyoma and its differentials, awareness of other benign vulvar tumors is important to provide best possible patient management. Tumors arising from the epidermis include hidradenomas, papillomas, and epidermal inclusion cysts. Those arising from the dermis are the lymphangiomas, the fibromas, and the lipomas. Occasionally, endometrial tissue may form in the pelvic diaphragm and the vulvar in cases of endometriosis.

Hidradenomas are rare tumors, arising from the apocrine sweat glands of the vulvar skin.[12] Vestibular papillomas are finger-like projections on the vulva and are easy to identify. Epidermal inclusion cysts are ingrowths of the epidermis into the dermis, forming a semi-solid cystic enclosure within the dermis, with average diameter of 2cm, lined by regular stratified squamous epithelium. The contents are mostly creamy keratin debris.

Cavernous lymphangiomas are rare, with only 10 reported in the literature.[13] Fibromas are solid tumours resulting from proliferation of connective tissue fibroblasts in the dermis. Lipomas arise from proliferation of adipocytes in the vulvar dermis.

Vulvar leiomyoma presents a diagnostic challenge clinically as what readily comes to mind on finding a painless vulvar swelling in the anatomic landmark of the Bartholin's gland is a Bartholin's gland cyst.[14] A photograph of the vulvar tumor before or during surgery was not considered because of a presumed diagnosis of a Bartholin's cyst, a commonly occurring vulvar lesion. Final diagnosis was made during histological study of sections of the excised tumor.

Early presentation of vulvar lesions, including tumors, should be emphasized to women, irrespective of absence of physical accompanying disabilities. About the only vulvar lesion that prompts women to hospital is Bartholin's disease, notably when complicated by infection. It is therefore not surprising that this patient presented rather late.

Early biopsy of any vulvar lesion or mass, is necessary, to confirm benign conditions or detect early, non-aggressive stages of malignant neoplasia.

Conflicts of Interest
None

Financial Support
None

Legends for Figures

Figure 1: Diagrammatic representation of right vulvar tumor pre-surgery

Figures 2a and 2b: Excised vulvar tumor Gross

Figures 3a and 3b: Excised vulvar tumor Cut surfaces

Figures 4a, 4b, and 4c: Excised vulvar tumor - Microscopy

Ethics
Approval was obtained from the Hospital Ethical Committee for this case report and review.

The Manuscript
This manuscript has been read and approved by all the authors. The requirements for authorship, in accordance with TJOG guidelines, have been met. Each author believes that the manuscript represents honest work.

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