

Squamous cell carcinoma of the Bartholin Gland: A rare genital cancer managed at the JFK Liberian-Japanese Friendship Maternity Hospital Monrovia, Liberia

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Abstract

Bartholin gland carcinoma is a rare tumor that constitutes 2-7% of all vulvar cancers. We presented a 40-years-old multipara who presented to the outpatient Gynecologic clinic of the JFK Maternity Hospital with a complaint of painful vulvar swelling of 3 years duration. The swelling began as a small lump on the left labium majus and grew overtime becoming increasingly painful, ulcerated with associated bleeding and malodorous discharge. Her last sexual activity was 2 years ago, of which she experienced dyspareunia but no post coital bleeding. Vulva examination revealed an ulcerated fungating lesion involving the left labium majus and minus, firm, tender, extending 2 cm into left vaginal wall and lower aspect of the right labium minus. The diagnosis of Bartholin gland carcinoma was made with Bartholin gland abscess and Vulvar cancer as differentials. Preoperative Fine Needle biopsy (FNA) showed acute on chronic inflammation of the Bartholin gland. A left hemivulvectomy was done and specimen was sent for histology, which revealed locally invasive squamous cell carcinoma of the Bartholin gland. Patient recovery was uneventful and she was discharged home 2 weeks post operatively on supplements and advised to follow up for chemotherapy.

Introduction

Primary Bartholin Glands Carcinoma (BGC) is exceedingly rare genital cancer and accounts for fewer than 5% of all vulvar carcinomas.¹ Criteria for the diagnosis of

BGC were first described by Honan in 1897 and subsequently revised by Chamlian and Taylor to include other features comprising of: the tumor involving the area of the Bartholin gland is histologically compatible with the origin from the Bartholin gland; areas of apparent transition from normal elements to neoplastic ones are found in histologic study; and there is no evidence of primary tumor elsewhere.² In 2018, the world literature search showed that there are currently 70 reported cases of Bartholin's gland carcinoma.^{3,4} In Liberia, this is the first reported case of Bartholin gland cancer. This shows how rare cancer of the Bartholin's glands is. Clinical presentation of primary BGC is usually late, lesions are deep within the vulva often misdiagnosed as a Bartholin's gland abscess or cyst.

Case Report

The patient was a 40-years-old Liberian Female, P5+1, with 4 living children. She attained tertiary level of education. She presented to the outpatient Gynecologic clinic of the JFK Maternity Hospital in June 2018. She had, as chief complaint, a painful vulvar swelling of 3 years duration. The swelling began as a small lump on the left labium majus and grew overtime becoming increasingly painful, ulcerated with associated bleeding and malodorous discharge. Her last sexual activity was 2 years ago, of which she experienced dyspareunia but no post coital bleeding. She had been prescribed several unspecified antibiotics and analgesics for treatment and relief of symptoms but no improvement. She was single, her six pregnancies were fathered by 3 partners, one of which died 10 years ago of unknown illness. Two of her previous partners were alive and well. Her menarche was unknown, her menses were regular, 3/28, no dysmenorrhea. There was no known chronic familiar illness. She was unemployed and had no history of consumption of alcohol or cigarette smoking.

On examination, she was in good general condition, but pale, no jaundice, no edema. No abnormal chest and abdominal findings. Vulva examination revealed an ulcerated fungating lesion involving the left labium majus and minus, firm, tender, extending 2 cm into left vaginal wall and lower aspect of the right labium minus. The rectum was free; cervix pink, uterus and adnexa findings were normal (Figure 1). The diagnosis of BGC was made with Bartholin gland abscess and vulvar cancer as differentials.

Her hematocrit level was 27%, HIV negative, she had normal liver function tests

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Availability of data and materials: All data underlying the findings are fully available.

Ethics approval and consent to participate: No ethical committee approval was required for this case report by the Department, because this article does not contain any studies with human participants or animals. Informed consent was obtained from the patient included in this study.

Consent for publication: The patient gave her written consent to use her personal data for the publication of this case report and any accompanying images.

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(LFTs) and kidney function tests (KFTs). Preoperative Fine needle biopsy (FNA) showed acute on chronic inflammation of the Bartholin gland. A left hemi-vulvectomy was done (Figure 2), and specimen (Figure 3) was sent for histology.

The surgical pathology specimen consisted of a hemi-vulvectomy specimen measuring 11cm length by 4cm width by 4cm depth. On the internal surface there was a firm mass measuring a length of 4cm by 2 cm width by 1cm depth. Cut section revealed firm tissue, lateral margin grossly free of tumor (Figure 3).

Shown in Figure 4 is the histopathological stain of sections from the specimen, which revealed locally invasive squamous cell carcinoma of the Bartholin's gland. Patient recovery was uneventful and she was discharged home 2 weeks post operatively on supplements and advised to follow up for chemotherapy.

Discussion

BGC is usually a slow growing tumour with a marked propensity for local invasion. Histologically Bartholin's gland carcinoma is classified into: adenocarcinoma, squamous cell carcinoma, adenoids cystic carcinoma, undifferentiated, adeno-squamous carcinoma.⁵ Most cases are squamous cell or adenocarcinoma.

Criteria for the diagnosis of BGC include: the tumour involving the area of the Bartholin gland, is histologically compatible with the origin from the Bartholin gland, areas of apparent transition from normal elements to neoplastic ones are found in histologic study and there is no evidence of primary tumour elsewhere.²

In this patient, the histology showed atypical squamous cells with cytoplasmic ringing, anisonucleosis with fine chromatin pattern in a background of inflammatory cells and necrotic debris consistent with squamous cell carcinoma of the Bartholin's gland.

Bartholin's gland carcinoma occurs typically around the middle of the 5th decade of life.⁶ Our patient was a little bit younger at the age of 40 years. Bhalwal *et al.*¹ also reported younger age at presentation of Bartholin's gland cancer. High level of education would have contributed to early presentation and diagnosis in our patient. Increasing rate of infections with Squamous cell carcinoma of the Bartholin's gland arises usually in the transition zone of the duct of Bartholin's gland, similar to the transition zone of the uterine cervix.⁷ It is associated with Human Papilloma Virus (HPV) infection.⁶ Our patient had multiple sexual partners.

The optimum treatment for this condition is still in debate, because of its rare nature many gynaecologists do not manage enough cases to gain wide clinical experience.

There is a tendency today to perform less radical surgery, hemi-vulvectomy or a wide local excision in order to obtain tumour clearance with negative margins.

The greatest difficulty in surgical management is with primary wound closure and healing and various techniques have been put to use.⁸ The incidence of wound breakdown and sepsis is also stated to be high. This difficulty was, however, not encountered with this patient. Hyperbaric oxygen therapy to improve wound healing has been recommended by some workers.⁹

Squamous cell carcinoma of the Bartholin's gland is a rare vulval carcinoma amongst women of reproductive age; correct diagnosis is often delayed because of focus on differential conditions such as

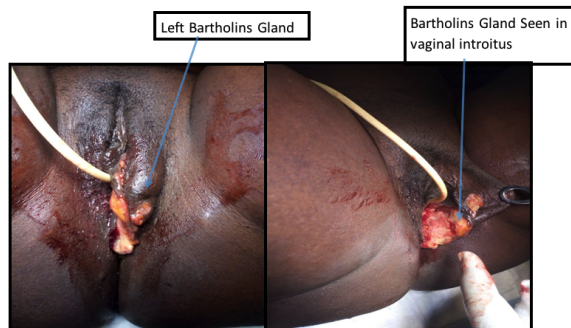


Figure 1. A) Carcinoma of the (L) Bartholin's gland with surface excoriation; B) carcinoma of the (L) Bartholin's gland visible swelling from the retracted introitus.

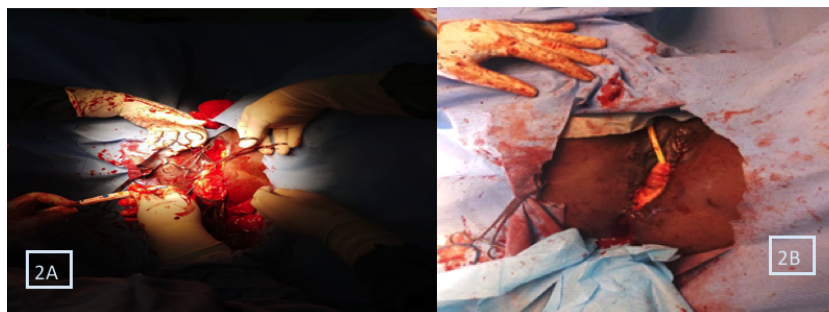


Figure 2. A) Left hemi-vulvectomy operation with wide excision of margins; B) re-constituted vulva.



Figure 3. Wide local excision flap – involving clear margin with the left Bartholin gland *in situ*.

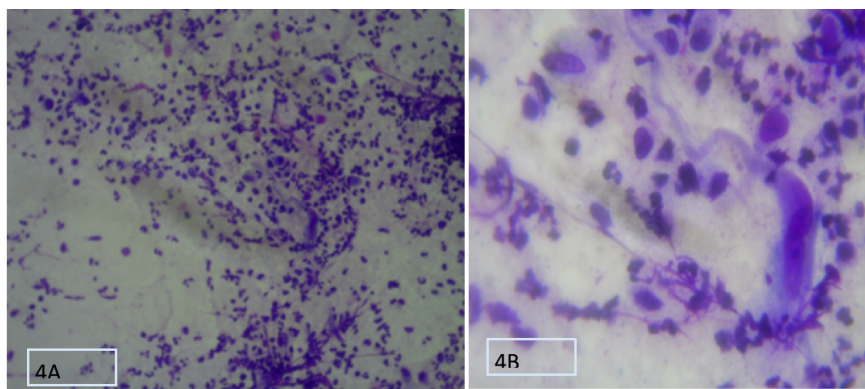


Figure 4. Histopathology: A) 4 X 10 Magnification; B) X 40 Magnification. Histopathological Report: Histology showed atypical squamous cells with cytoplasmic ringing, anisonucleosis with fine chromatin pattern in a background of inflammatory cells and necrotic debris consistent with squamous cell carcinoma of the Bartholin gland, locally invasive.

Bartholin's gland cysts and abscesses.

When asymptomatic cysts become enlarged or infected and cause significant pain, physicians are tempted simply to undertake incision and drainage or marsupialization.

We recommend that specimens should be taken for histology and the patient screened for human papilloma virus if tests are available.

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