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Total Radical Vulvectomy and Bilateral Inguinofemoral Lymphadenectomy for Vulva Carcinoma: Case Report and Review of Literature

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Abstract

Introduction: Vulva cancer is an uncommon disease. It represents about 3% to 5% of gynecologic cancers worldwide, but its incidence is increasing with age and Human Papilloma Virus (HPV) and HIV infection. In Cameroon, annual number of new cases was 52 in 2020. Surgery is the treatment of choice and it is only realized in teaching hospitals. Yaounde Gynaeco-Obstetric and Pediatric Hospital (YGOPH) is one of the centers where cancer cases are referred to. **Case Report:** We report a case of a 49-year-old woman, gravida 2 para 1, known HIV positive for twenty years, who complained of vulva pruritis for last two years. Six months after the onset of pruritis, she presented a local swelling of both labia majora, which got ulcerated on the right, associated with severe pain. On clinical examination, she was in good general condition with a 3 × 3.5 cm ulcerated, budding mass in the middle third of the right labia majora, approaching the midline formed by the vaginal orifice, with no locoregional infiltration (vagina, bladder, urethra, anus and rectum). The left labia majora was abnormally thickened by about 1 cm, with no palpable mass. There were no palpable inguinal lymph nodes. Punch biopsy revealed an invasive non keratinizing carcinoma of the vulva. MRI showed no suspicious

lymph nodes or invasion of pelvic organs. The patient underwent radical vulvectomy with bilateral inguinofemoral lymph nodes dissection. The postoperative period was uneventful, and anatomopathological examination confirmed vulvar carcinoma without lymph node invasion or lymphovascular emboli, and excision margins were in sano. No adjuvant treatment was therefore initiated. **Conclusion:** Vulva cancer is extremely rare, and frequently associated with HIV infection. Any suspected vulvar lesion should be biopsied. Although other treatment modalities are available, surgery remains the cornerstone of its management.

Keywords

Vulva, Cancer, Radical Vulvectomy, Inguinofemoral Lymphadenectomy, Cameroon

1. Introduction

Vulvar cancers are an extremely rare disease, ranking as the 29th most common cancer, with an incidence of 47336 cases in 2020 [1]. It is the fourth gynecologic malignancy, accounting for 3% to 5% of all malignant neoplasms of the genital tract [2]. There are few data on vulvar cancer in Cameroonian women with approximately 52 new cases in 2020 [3]. In earlier studies conducted in Cameroon, vulva cancer accounted for 0.91% to 4% [4] [5].

It occurs mainly in postmenopausal women over 55 years old [6], even if some cases have been described before 30 years old [7]. Other risk factors associated with vulvar cancer are smoking, HIV, lichen sclerosus, HPV and vulvar intraepithelial neoplasia (VIN). In developing countries, around 60% of vulvar cancers are associated with HIV and HPV infection [8]. Similarly, to cervical cancer, oncogenic HPV (16, 18, 31, 33, 66) may contribute to the appearance of vulvar cancer, as well as cancer of vagina, anus, penis and larynx [9]. For VIN, the terms VIN1, VIN2 and VIN3 are no longer used, and are simply called VIN [10]. However, there are two types of premalignant vulvar lesions: HPV-induced intraepithelial neoplasia (vulvar high grade squamous intraepithelial lesions or Vu-HSIL, also known as usual vulvar intraepithelial neoplasia or uVIN, and differentiated vulvar intraepithelial neoplasia or dVIN [11]. uVIN is often caused by HPV infection, and occurs more frequently in younger patients. The risk of malignant transformation is estimated at 5.7% [12]. However, dVIN develops from epithelial lesions responsible for chronic inflammation or vulvar lichen, and occurs in older patients [13]. The risk of malignant transformation is high, estimated at 32.8% [12].

Approximately 70% of vulvar cancers are squamous cell carcinomas. The basaloid and verrucous forms account for a third of cases, and develop from Vu-HSIL; the keratinized form readily develops from epithelial lesions responsible for chronic inflammation or vulvar lichen responsible for dVIN [14]. Other less frequently histological types are melanoma, adenocarcinoma, extramammary Pa-

get's disease, sarcoma.

Diagnosis of vulvar cancer is based on clinical gynecological examination and biopsy. Vulva pruritis is frequently found [15], but clinical manifestations are highly variable and non-specific. Other symptoms include plaques, ulcerations, masses, bleeding, urinary symptoms such as dysuria or palpable adenopathies, particularly inguinal [16].

The management of patients with vulvar cancer is well codified by European Society of Gynaecological Oncology (ESGO) guidelines updated in 2023 [17], and depends on the staging of the disease. The most recent revision to vulvar cancer staging by the International Federation of Gynecology and Obstetrics (FIGO) Committee for Gynecologic Oncology was made in 2021 [18]. However, the 2017 8th version of TNM staging of vulvar cancer can also be used [19].

The aim of this work was to demonstrate the surgical aspects of a radical vulvectomy with inguinofemoral lymph node dissection in a patient with stage IB vulvar cancer managed in YGOPH in Cameroon.

2. Case Presentation

A 49-year-old woman, gravida 2 para 1, known HIV positive for twenty years, complained of vulva pruritis for last two years. Six months after the onset of pruritis, she presented a local swelling of both labia majora, which get ulcerated on the right, associated with severe pain.

On clinical examination, she was in good general condition with a 3 × 3.5 cm ulcerated (**Figure 1**), budding mass in the middle third of the right labia majora, approaching the midline formed by the vaginal orifice, with no locoregional infiltration (vagina, bladder, urethra, anus and rectum). The cervix was absent because she underwent a total hysterectomy 15 years earlier for abnormal uterine bleeding due to leiomyoma. The left labia majora was abnormally thickened by about 1 cm, with no palpable mass. There were no palpable inguinal lymph nodes. Punch biopsy revealed an invasive non keratinizing carcinoma of the vulva. MRI showed no suspicious lymph nodes or invasion of pelvic organs. After discussion at a multidisciplinary consultation meeting, the patient was classified as stage 1b, psychological support was provided and primary surgery was recommended. We preferred radical vulvectomy to wide excision, because postoperative follow-up is often difficult due to patient's lack of financial resources. In the absence of sentinel node technology in our setting, we performed bilateral inguinofemoral lymph nodes dissection.

The vulvectomy procedure was performed as follows:

Incision: A butterfly wing incision was made over the vulva, with a 2 cm margin of normal tissue around the tumor in all direction. A 1cm margin was left around the urethral meatus, clitoris and anus (**Figure 2**).

Main dissection: The labiocrural incisions were carried through the fatty tissue bilaterally until the level of the deep fascia of the urogenital diaphragm. The internal pudendal vessels were individualized, clamped, transected, and ligated.

Caudally, the perineal body and posterior vulvar tissue were dissected from the anus. Cranially, the specimen was dissected off the pubic periosteum. Laterally, the dissection was taken deeply until we encountered the adductor fascia. The clitoris, vestibule and urethral meatus have been preserved. The portion of the vulva tissue along the perineum are dissected upward to the vagina. The vascular vestibular tissue along the vagina was clamped and transected. The specimen was now free cranially and caudally, and the removal of the vulva was done (**Figure 3**). A tension-free closure was done to close the edges of the wound and the perineum (**Figure 4**).

We then proceeded with bilateral inguinofemoral lymph node dissection as follows:

Skin incision: An elliptical incision was done parallel to and approximately 1 cm below the inguinal ligament from the anterosuperior iliac spine to the pubic tubercle.

Main dissection: This procedure is centered on the triangle of Scarpa, which is defined as follows: Cranially by the inguinal ligament composed by the aponeurosis of external oblique muscle and located between the anterior superior iliac spine and the pubic tubercle, laterally by the Sartorius muscle and medially by the adductor longus muscle;

The surgical procedure begins by performing a 6 to 8 cm incision parallel and superior to inguinal ligament. The incision of the subcutaneous fatty tissue must be perpendicular to the plane of the skin incision, not to remove the subcutaneous tissue, which could cause skin necrosis. Until reaching the superficial Camper's fascia, which is the superficial limit of the superficial lymphadenectomy.

Next, we will find the inguinal ligament. If the cutaneous incision is performed above the inguinal fold, the first anatomical structure that we will find below Camper's fascia is the inguinal ligament. The lymph node should be dissected from the inguinal ligament. Starting at the upper edge and progressing downward toward the femoral triangle.

The superficial lymph node should be dissected circumferentially from the surrounding subcutaneous fatty tissue. While removing superficial lymph node, it is important to identify the superficial vessels such as the circumflex and epigastric vein and section them distal at this point.

As we proceed toward deeper structures, we will come across the cribiform fascia, which is an extremely thin and multiperforated membrane that can be easily penetrated.

At this level, the surgeon must perform a meticulous dissection to prevent injuring the saphenous vein which perforate the cribiform fascia at this level. Even if the saphenous vein can be ligated, it is preferable to preserve it to minimize the risk of lymphedema.

Once the saphenous vein has been identified and the cribiform fascia incised, the superficial lymph node can be removed. At this point we proceed to seal and cut the proximal part of the circumflex vein on its entrance into the saphenous vein.

After the incision of the cribiform fascia and the removal of superficial lymph node, the deep territory become visible. The deep lymph nodes are located medially to the saphenous and femoral veins, and laterally to the adductor longus muscle. It is not required to visualize the apex of the Scapa triangle but only the first 3 cm from the entry of the saphenous vein into the femoral vein. The deep lymphadenectomy should not include the lymph node situated laterally to the femoral vein.

Using a vessel sealer to perform the dissection and removal of the tissue, can be beneficial to seal the small lymphatic vessels and to reduce the risk of postoperative lymphocele. After removal of the deep node, the surgeon is able to identify the floor of the dissection: pectineus muscle, the femoral and saphenous veins and the femoral fascia. At the end of the procedure, we can identify the femoral vein, the saphenous vein, the external pudendal vein and the stump of the circumflex and epigastric vein. We can also identify the pectineus muscle and the lateral border of the adductor longus muscle which can be Feld but not seen.

It should be noted that the femoral artery and nerve are not visible during the lymphadenectomy because they are covered by the femoral fascia and therefor, the Sartorius muscle is not visible as well.

The postoperative period was uneventful, and anatomopathological examination confirmed vulvar carcinoma without lymph node invasion or lymphovascular emboli, and excision margins were in sano. Unfortunately, we don't have the technology to provide a microscopic picture of the histology. No adjuvant treatment was therefore initiated. The patient has been counselled on the risk of recurrence. Post-operative follow-up will take place over 5 years and should include, at a minimum, a symptom review and a complete physical examination of the vulva, skin and inguinofemoral lymph nodes, and if necessary, imaging and laboratory test may also be requested (**Figure 5** & **Figure 6**).

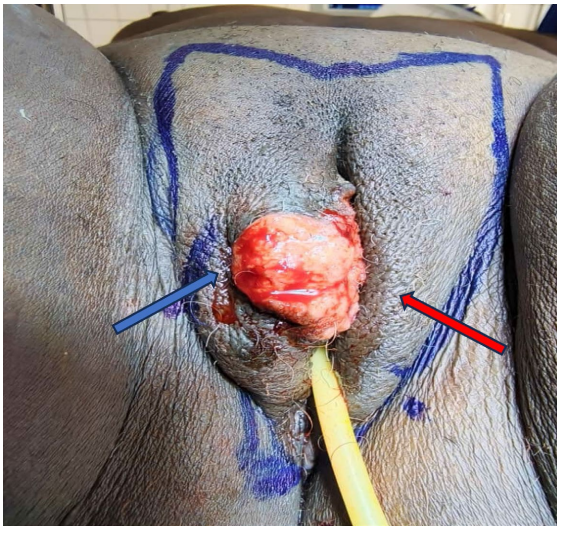

Figure 1. Non-lateralized right labia majora ulcerated cancer of about 3 × 3.5 cm (blue arrow) with thickening of left labia majora (red arrow). Ink mark on skin before incision.

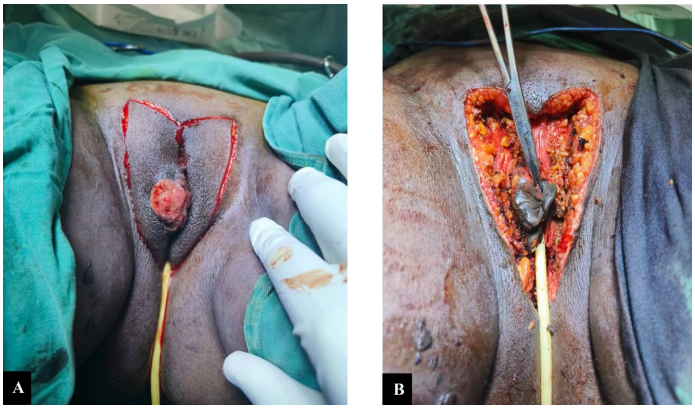

Figure 2. (A) Skin incision surrounding both the mass of the right labia majora and the thickening of the left labia majora; (B) Aspect of the primary site after radical vulvectomy.

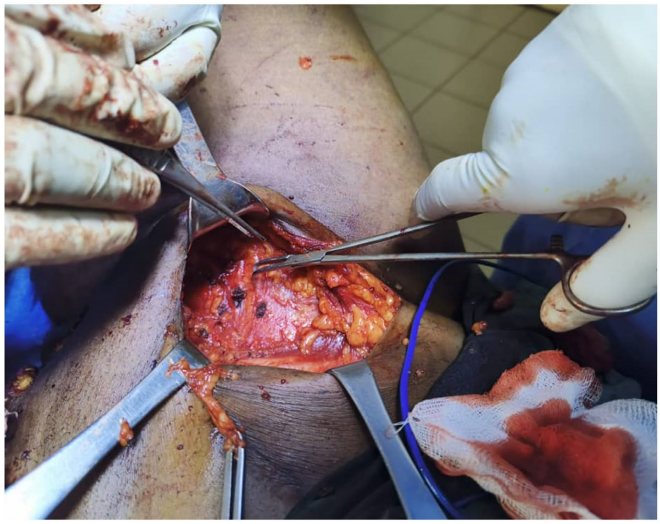

Figure 3. Aspect of right inguinofemoral lymph node dissection.

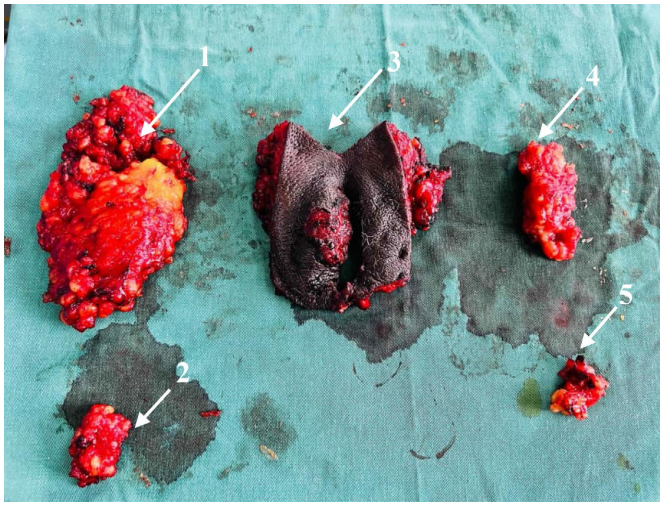

Figure 4. (1) Right superficial inguinofemoral lymph node; (2) Right deep inguinofemoral lymph node; (3) Aspect of resected primary tumor; (4) Left superficial inguinofemoral lymph node; (5) Left deep inguinofemoral lymph node.

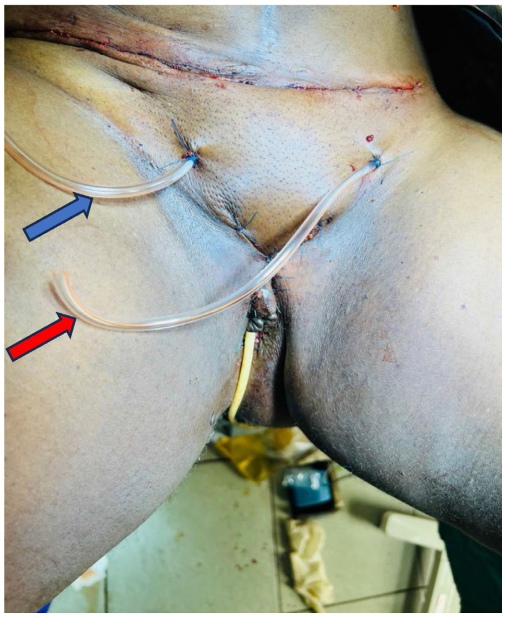

Figure 5. Final view after Surgery. Drain for lymphatic fluid collection. Right inguinofemoral drainage (blue arrow) and left inguinofemoral drainage (red arrow).

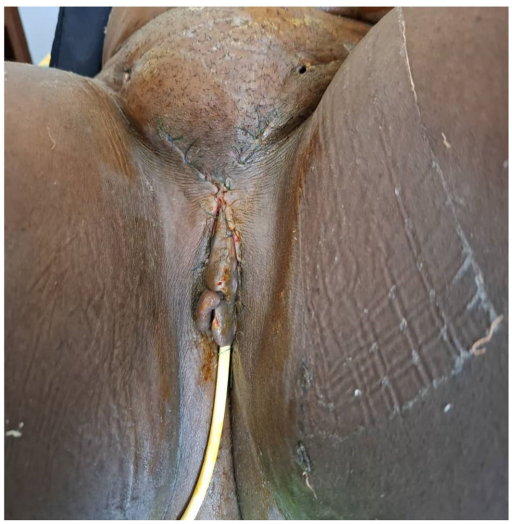

Figure 6. Satisfactory cosmetic appearance. Day 10 after surgery.

3. Discussion

Although the incidence of vulvar cancer has doubled over the last 15 years, especially in young women aged between 30 and 49, it remains a rare disease [20] [21], representing the 4th gynecologic malignancy, accounting for 3% to 5% of all malignant neoplasms of the genital tract [2]. The identified risk factor in our case was the HIV status of our patient. Our patient had not been tested for HPV, but it should be remembered that 60% of vulvar cancers are associated with HPV infection in developing countries [8]. Our patient had not yet reached menopause, although vulvar cancer does preferentially affect postmenopausal women [6]. However, some cases have been described in patients under the age of 30 and would

probably be HPV-induced [7]. In young patients, HPV can induce premalignant lesions called vulvar high grade squamous intraepithelial lesions or Vu-HSIL, also known as usual vulvar intraepithelial neoplasia or uVIN [11], with 5.7% risk of malignant transformation [12]. As these precancerous lesions are not visible, they are more likely to be diagnosed in women with risk factors who have local vulvar signs. Unfortunately, in our case, as in most cases in developing countries, women are diagnosed very late when the cancer has already set in [22]. Clinical manifestations are not specific. Our patient presented with vulvar pruritis. This symptom is frequently found in the literature [15].

Surgery is the cornerstone for staging and treatment of vulvar cancer, and includes both primary tumor resection and inguinofemoral lymph node dissection. The staging is very important for the management of the disease, and both the 2021 FIGO classification [18], and the 2017 8th TNM classification system [19] can be used. In the past, "en bloc" resection was considered the gold standard [23]. This mutilating technique, popularized by Taussig [24] and Way [25], consisted of a radical vulvectomy with "en bloc" bilateral pelvic and inguinofemoral lymphadenectomy. Today, the vulvar cancer surgery has been demoted from "en bloc" complete radical vulvectomy to radical local excision with tumor-free margins of at least 8mm, even if the evidence of this cut off is low [16]. The margin of 1cm is recommended by some and if the tumor is in close proximity to the clitoris or urethra, smaller margins may be considered in an attempt to preserve their function [17]. In our case, as the patient complained of bilateral pruritus, with thickening of the contralateral labia majora, we preferred to make a bilateral excision with a margin of 2 cm on the outside and 1 cm on the inside, as the lesion was slightly in midline (**Figure 3**). If the margins are close (less than 8mm) or are not in sano, a second resection should be attempted. De Hullu *et al.* [26] and Chan *et al.* [27] showed no local recurrence when pathologic margins were greater than 8 mm in microscopy, corresponding to macroscopic measurement for about 2cm. Adjuvant local radiotherapy is recommended if persistent positive margins or if the patient is not eligible for a second operation [17]. In cases of multifocal invasive disease, radical excision of each lesion as a separate entity may be considered. When these multifocal invasive lesions have as background an extensive vulvar dermatosis, vulvectomy may be indicated [17].

Lymph node assessment is an important prognostic factor in vulvar cancer, as well as size of primary lesion, depth of invasion, and involvement of the lymphovascular space [28]. Only 25% - 30% of cases are presenting inguinal lymph node metastasis; therefore, overtreatment of patient will be frequent [23]-[30]. Several complications associated to inguinofemoral lymphadenectomy are lymphedema of the lower extremity (14% - 49%), lymphocyte formation (11% - 40%) and wound infections with dehiscence [31]. Due to lack of Sentinel Lymph Node (SLN) technique in our hospital, we systematically performed a bilateral inguinofemoral lymphadenectomy on our patient. Over the few decades, SLN dissection for early vulva cancer has been shown to be safe, accurate and cost effective, making it the most preferred

approach to lymph node assessment. This procedure is recommended in patient with unifocal cancers of < 4 cm, > T1a, without suspicious inguinofemoral nodes [17]. For non-lateralized (less than 1 cm from the midline) or central tumor, as lymphatic drainage is often bilateral, the SLN dissection should be taken bilaterally. The incidence of disease recurrence is 10.5% in patient with multifocal lesion, higher than the 2.3% of recurrence rate of those with unifocal lesion, therefore, SLN dissection is not recommended in case of multifocal tumor [32]. False-negative are possible with SLN biopsy, especially in the presence of positive groin metastasis which can obliterate the lymph flow and cause bypassing of the sentinel node [33]. In case of SLN failure (SLN not found), inguinofemoral lymphadenectomy should be performed [17]. It is mandatory to perform at least ipsilateral inguinofemoral lymphadenectomy by separate incisions for tumors ≥4 cm and/or in case of multifocal lesion, and when this lymphadenectomy has demonstrated metastasis, contralateral inguinofemoral lymphadenectomy may be performed. Ipsilateral inguinofemoral lymphadenectomy should be performed if macrometastasis (> 2 mm) are found during the SLN procedure. When micrometastasis (≤ 2 mm) or isolated tumor cells are found in the metastatic SLN, adjuvant treatment by radiotherapy should be preferred to inguinofemoral lymphadenectomy [17].

In order to reduce the risk of local recurrence, which occurs in 40% - 50% of cases after surgery [34], adjuvant chemoradiotherapy is often necessary, although this is controversial in vulvar cancer [35]. Adjuvant radiotherapy of the vulva is recommended in various situations including positive margin where re-excision is not possible, close margins, lymphovascular or perineal invasion, large tumor size, and/or depth of invasion > 5 mm [17] [34] [36]. For the inguinofemoral region, postoperative radiotherapy is indicated when more than one metastatic lymph node and/or extracapsular spread are identified after inguinofemoral lymphadenectomy [17]. As all these risk factors for local recurrence were absent in our patient, she did not receive adjuvant treatment.

Treatment of advanced vulvar cancer involves several treatment modalities, including surgery, radiotherapy and chemotherapy, and should be considered after a multidisciplinary consultation assessment [37] [38]. The use of targeted therapeutics in the management of advanced vulva cancer is still unknown [39] [40].

4. Conclusion

Vulvar cancer is an extremely rare gynecological disease. Surgery remains the cornerstone of its management even if it has been demoted from radical vulvectomy to wide local excision. In developing countries, in the absence of the sentinel lymph node and patients' limited financial resources to carry out preoperative workup and follow-up workup, it is understandable to be radical during the surgery.

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Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this article.

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