

# **Extensive Vulvectomy and Reconstructive Flaps for Advanced Vulvar Cancer in Young Woman: Case Reports**

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#### ABSTRACT

**Introduction:** Vulvar cancer is a rare disease accounting for approximately 5% of female genital tract tumors worldwide. It is a squamous cell cancer that mostly affects older women over the age of 65. In the existing body of literature, no instances of vulvar cancer have been documented in the form of case reports. Therefore, this study aimed to report the challenges and outcomes of vulvectomy with reconstructive flap in vulvar cancer survivors in a 39-year-old woman.

Case Presentation: A 39-year-old woman with a palpable lump around the vulva was present and being examined. The anatomic pathology report (APR) features suggested poorly differentiated malignant tumors. A neoadjuvant chemotherapy was then administered to the patient aimed at shrinking the size of the tumor. However, there was no effective response to the chemotherapy, which resulted in a progressive tumor. Extensive vulvectomy and reconstructive flap were carried out to support quality of life, and the outcomes were reported, including flap, survival, complications, and mortality. These outcomes were evaluated between November 2021 and March 2023. No total flap loss was observed, and neither was there any donor site morbidity or mortality during the process.

**Conclusions:** This study showed that vulvar cancer was a rare condition at a young age. Advanced vulvar cancer was found in the patients and extensive vulvectomy surgery was carried out followed by a pedicled ALT flap. This option was currently the best for immediate reconstruction due to the preservation of sensibility and tissue availability in the donor areas. The result showed no wound dehiscence, marginal necrosis, or surgical site infection. After 15 months of follow-up, the flap was observed to have good vascularization. The association of the Gynecologist with the Plastic Surgeon offered palliative care to improve the quality of life of the patient and provide good postoperative results.

### **INTRODUCTION**

Vulvar cancer is a rare disease accounting for approximately 5% of female genital tract tumors worldwide. According to Globocan, the estimated number of women diagnosed with this disease in 2020 is 42,240. This cancer accounts for approximately 2–4% of malignant tumor cases in the lower genital tract, affecting two of every 100,000 women in developing countries [1]. It is a rare condition affecting older women and accounts for 3–5% of all gynecological cancers. The increased age is itself a high-risk factor, with the most common symptoms being pruritus, ulcer, vaginal discharge, or pain [2].

Diagnosis is inferred by medical history, clinical examinations, vulvar biopsy, and diagnostic imaging [3]. The most commonly reported histologic type of tumor is squamous cell carcinoma, accounting for 96%, with survival of about 71.9% [4]. Two distinctive pathways are recognised, driven by chronic inflammation, primarily lichen sclerosus lesions (LS), or high-risk Human Papillomavirus (HR-HPV). According to Schnack et al. [5], the disease is diagnosed in early stages (stage I or II) confined to the vulva in about 60 to 70% of the cases. The staging of vulvar cancer is popularly carried out according to the International Federation of Gynecology and Obstetrics (FIGO). It is curable when diagnosed at an early stage, and survival is largely due to the absence

of lymphatic metastases. Lymph node positivity is an independent bad prognostic factor [4].

The initial stage of the disease is treated surgically, while radiotherapy and chemoradiotherapy are used in advanced stages [4]. Although primary vulvovaginal reconstruction has a high potential to improve the outcome of patients after vulvectomy, flap reconstruction is not an established part of the current standard treatment for vulvar cancer. According to a previous study, vulvar cancer often requires radical vulvectomy with subsequent vulvar flap [6].

Approximately in 20 to 60% of cases, there are postoperative complications ranging from infection to flap necrosis that often require reoperation [7]. A previous study reported postoperative dehiscence, lymphocysts, and lymphedema rates as high as 64 to 85% [8]. Due to the lack of cases reported and the challenges that may be encountered specifically at young age, this study aimed to provide palliative care to young women with advanced vulvar cancer.

### **CASE PRESENTATION**

A 39-year-old woman was referred from Karawang Public Hospital to the emergency installation in Dharmais Cancer Hospital with a chief complaint of lump and wart-like growth on the vulva, accompanied by persistent pain and soreness.

On 23<sup>rd</sup> March 2021, magnetic resonance imaging (MRI) of the abdomen and pelvis showed an inhomogeneous mass lesion on the vulva with multiple

nodulars over the right and left labia majora and perineum. The imaging also showed bilateral inguinal and left obturator lymphadenopathy, a 3.8 x 4.37 cm mass on the area of the vulva with multiple nodular lesions with a diameter of 0.9–2.6 cm, and multiple nodular lesions on the perineum with a diameter of 1.3–2 cm. Other characteristics shown are right and left inguinal lymphadenopathy with diameters of 0.8 to 3.6 cm and 0.8–2.3 cm, respectively, as well as left obturator lymphadenopathy with 0.77 cm (Figure 1A). In addition, confirmation by immunohistochemistry (IHC) test of the vulva and fine needle aspiration biopsy of the inguinal showed large cell neuroendocrine carcinoma. This shows that the diagnosis of the patient was right vulvar cancer.

On 30th August 2021, the patient immediately began receiving neoadjuvant chemotherapy with an etoposide cisplatin II (200/70) regimen for 3 cycles. However, a progressive inhomogeneous mass was developed around the vulva after receiving neoadjuvant chemotherapy.

On 11<sup>th</sup> October 2021, multislice computerized tomography (MSCT) of the abdomen and pelvis showed a 5 x 4.8 cm mass, which was formerly 3.8 x 4.37 cm, on the vulva area diameter of 1.3–3.2 cm. The tomography also showed multiple nodular lesions on the perineum, right, and left inguinal lymphadenopathy, with diameters of 1.8–4.6 cm, 0.9–5.3 cm, and 1–3.4 cm, respectively. It also showed left obturator lymphadenopathy with a diameter of 1.2 cm, formerly 0.77 cm, as shown in **Figure 1B**.

A decision was made after the Tumor Boards (TBs) meeting, including the team of gynecology oncology,









Figure 1. Vulva lesion progress. (A). Primary vulva lesion (lump and wart-like growth on the vulva) at presentation to the hospital; (B). Progressive vulva lesion after 3 cycle neoadjuvant chemotherapy; (C). Wide extensive vulvectomy and inguinal lymphadenectomy; (D). Pedicled anterolateral thigh (ALT) flap reconstruction after wide extensive vulvectomy and inguinal lymphadenectomy

reconstructive plastic surgery, radiation oncology, medical oncology, palliative medicine, and anesthesiologist. The decision was to administer the best multidisciplinary treatment plan and integrated palliative care to improve the patient's quality of life. Due to the large defection, vulvar reconstruction was performed 1 month later.

On 19th November 2021, the patient underwent a wide extensive vulvectomy and inguinal lymphadenectomy followed by pedicled ALT flap reconstruction, as shown in **Figure 1C**. The histopathology examination showed neuroendocrine carcinoma, and the patient was given preoperative deep vein thrombosis prophylaxis with heparin, as well as preoperative antibiotics with cefazolin. The patient was placed in a dorsal lithotomy position, prepped with chlorhexidine and betadine, and a Foley catheter was placed. A multidisciplinary surgical method was carried out including a gynecologic oncologist and plastic reconstructive surgeon to perform an extensive vulvectomy, followed by a reconstructive flap.

## Patients treated with extensive vulvectomy and inguinal lymphadenectomy using en bloc method

Pedicled ALT flap reconstruction was carried out by a plastic reconstructive surgeon (Figure 1D). The defect on the pubic and vulva region measures 17 x 19 cm and extensive resection was carried out around 2 cm from the edge of the tumor. The type of flap is determined by the size and location of the defect. Flap harvest started with a medial incision, initiated and extended until reaching the deep fascia above the rectus femoris muscle. This process was continued until the full elevation of the flap was achieved. The perforator and pedicled ALT were skeletonized along their courses to the superficial femoral artery and measured. Dissection of the flap was completed with 2 perforators and the ALT flap was tunneled beneath the rectus femoris muscle required for a tension-free inset. It was then rotated to varying degrees and inset into the defect after ensuring viability and rechecking the vascularity while in the desired position. A drain is placed in the donor site as it is closed.

# Flap and secondary defect on the right medial thigh closed with 2-0 vicryl sutures and skin staplers

Split thickness skin graft (STSG) was harvested from the right thigh region and inset on the secondary defect, then sutured with 4-0 vicryl and skin staplers. Final defects were covered with tulle and a dressing on STSG was derived using elastic rubber bands to tie over the skin graft. Bandaging was soft, to avoid compression over flap, and the limb was held in an elevated position. A window was left uncovered for monitoring of color and temperature without bandage removal. The donor site/secondary defect in the patient was grafted andflap

was monitored post-operative. Furthermore, the parameters monitored included color, warmth, margins, signs of poor perfusion/congestion, epidermal shrinking, and blistering. The amount of blood lost reported during the surgical operation was 500 cc.

The postoperative wound was dressed with lidocaine jelly and anticoagulant gel every 4 hours and sealed with occlusive dressing. On the opening of the wound, drains were inserted for seven days. Serous hemorrhagic drainage occurred in the first 3 days after surgery with 70cc per 24 hours. The warming lamp vertically irradiated 40 cm from the flap for 3 days. No major intraoperative or postoperative complications occurred, and the patient was discharged within two weeks with no readmissions for wound care. The anatomic pathology test of the postoperative vulva and fine needle aspiration biopsy of the inguinal still showed large neuroendocrine carcinoma (Figure 2).

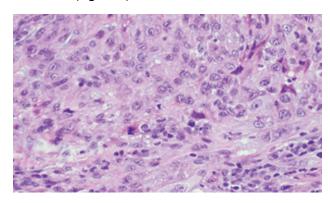


Figure 2. Revealed large cell neuroendocrine carcinoma



Figure 3. Evidence of recurrence on the donor site

The outcomes reviewed were flap survival, postoperative complications, donor site morbidity, and mortality. On the other hand, the postoperative complications observed include infection, dehiscence, arterial thrombosis, vein thrombosis, abscess formation, hematoma, bleeding, partial flap necrosis, and total flap loss.

On 4 January 2022, the patient had a common postoperative complication, such as postoperative

urinary retention followed by catheterization. After 15 months of follow-up, there was evidence of recurrence on the donor site, as shown in **Figure 3**. On 10<sup>th</sup> March 2023, the patient underwent a biopsy of the recurrence tumor and the anatomic pathology test showed large cell neuroendocrine carcinoma. The data obtained in this case report was gathered from medical records and the follow-up period ranged from November 2021 to March 2023.

### **DISCUSSION**

Vulvar cancer is a rare type with a characteristic of strong invasion, rapid progression, and easy recurrence [9]. Its staging is determined according to the American Joint Committee on Cancer (TNM) and the International Federation of Gynecology and Obstetrics staging systems [10]. From this case report, after receiving neoadjuvant chemotherapy, the tumor grows more rapidly than usual. This disease usually occurs in postmenopausal women, and some cases of young patients were also reported.

Vulvar cancer can be associated with troublesome symptoms, including pain, bleeding, and severe patient discomfort with consequent worsening of the quality of life (QoL), as well as psychosocial well-being and social isolation. Patients with incurable diseases can be treated with palliative care to relieve pain and other distressing symptoms [11]. Traditionally, oncologists focused their efforts on maximizing the overall survival rate of their patients. Recently, well-being and QoL considerations have become particularly important in gynecological oncology. In this case, patients are given important information showing that they retain the ability to urinate and defecate, but no longer have the capacity for sexual intercourse. Women submitted to surgical treatment for vulvar cancer may have difficulty in accepting their physical appearance, as well as coping with their urinary and sexual disorders. In this particular case, the patient experienced significant preoperative anxiety. According to de Melo Ferreira et al. [12], studies regarding QoL outcomes in women who survive vulvar cancer are still limited.

There is a critical quandary to consider in advanced vulvar cancer management. This is due to the severe negative impact of demolitive surgery on women who are afflicted by both functional and psychological consequences of the procedure [13]. Treatment of vulvar cancer depends primarily on histology and surgical staging. It is predominantly surgical, particularly for squamous cell carcinoma, although concurrent chemoradiation is an effective alternative, particularly for advanced tumors. Furthermore, the management of women with advanced vulvar cancer is complex and should be individualized and carried out by a multidisciplinary team [14]. Tagliaferri et al. [15] Reported that the multidisciplinary management of

advanced vulvar cancer patients, based on TBs discussion, represents the broader and more fruitful cooperation possible when choosing the best treatment for each patient. According to Specchia et al. [16], over the last decades, scientific evidence shows that cancer care has been increasingly delivered through multidisciplinary team (MDT) interventions. This case is also being handled by an MDT from the hospital including members of pathology and radiology to confirm whether the tumor can be excised with free margin by the gynecologic oncology department.

The treatment of vulvar cancer uses extensive vulvectomy or inguinal lymphadenectomy, which often includes resection of at least 2–3 cm around cancer foci, effectively reducing the local recurrence rate. Vulvar reconstruction is critical for cosmetic, functional, and psychological reasons. On the other hand, flap-based reconstruction is recommended for the treatment, including both primary or recurrent cases, and addressing early or late large lesions. ALT flap can be considered to be one of the workhorses of perforator flap. Its ease of dissection together with the long and reliable pedicle make it extremely suitable as a free-skin flap. According to Landuyt et al. [17], ALT flaps have been introduced in breast and genital, as well as upper and lower extremity reconstruction.

Recurrent vulvar cancer occurs in an average of 24% of cases after primary treatment, which may include surgery with or without radiation [18]. Most recurrences, accounting for 40–80% occur within 2 years after initial treatment. Surgery is still the main treatment modality for vulvar cancer, but radiation therapy also has an important role in management [19]. Radiation therapy is typically delivered either as an adjuvant to surgery or as a definite modality in conjunction with chemotherapy. After 15 months of follow-up, there was evidence of recurrence on the donor site due to the delayed adjuvant radiation therapy. On 17<sup>th</sup> March 2023, the patient underwent a biopsy of the recurrence tumor and the anatomic pathology test showed large cell neuroendocrine carcinoma.

Another retrospective study compared 77 patients treated with direct closure with 72 treated with reconstructive procedures. The result of the study showed that skin flap reconstruction decreases postoperative morbidity and provides better anatomical and functional results than direct closure of the perineal defect. Reconstructive operations reduced wound dehiscence, vaginal introitus stenosis, sexual dysfunction, and urine stream redirection to 26%, 2%, 10%, and 1%, respectively. These values were compared to 64%, 8%, 50%, and 5%, respectively, in the primary closure group according to the same study [20].

Early reduction of the occurrence of splitting and promotion of wound healing is beneficial for comprehensive treatment, such as radiotherapy, chemotherapy, and targeted therapy, which can be performed early after surgery. In this case, neuroendocrine surgery should be carried out, followed by adjuvant chemotherapy and radiation therapy. Unfortunately, the adjuvant chemotherapy and radiation therapy were delayed.

### **CONCLUSIONS**

This report showed that vulvar cancer was a rare condition at young age. Advanced vulvar cancer was found in the patients and extensive vulvectomy surgery was carried out followed by a pedicled ALT flap. This option was currently the best for immediate reconstruction due to the preservation of sensibility and tissue availability in the donor areas. The result showed no wound dehiscence, marginal necrosis, or surgical site infection. After 15 months of follow-up, the flap was observed to have good vascularization. The association of the Gynecologist with the Plastic Surgeon offered palliative care to improve the quality of life of the patient and provide good postoperative results.

### **DECLARATIONS**

### **Competing of Interest**

The authors declare no competing interest in this study.

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