

A Case of Erythroplasia of Queyrat Complicated with Verrucous Carcinoma

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Abstract Erythroplasia of Queyrat is a rare squamous cell carcinoma that occurs in middle-aged, uncircumcised men, and usually involves the glans. The cause of this disease is still unclear, although it may be related to a redundant prepuce, phimosis, and human papillomavirus infection, and may develop into squamous cell carcinoma. A case of Erythroplasia of Queyrat, complicated by verrucous carcinoma, is reported in this paper.

Keywords: Erythroplasia of Queyrat, squamous cell carcinoma in situ, verrucous cancer

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1. Introduction

Erythroplasia of Queyrat (EQ) and verrucous carcinoma of the penis are both low-grade malignant squamous cell carcinomas in situ, which are rare in the clinic. They typically occur in middle-aged, uncircumcised men. The causes and pathogenesis of both conditions are still unclear. However, the disorders may be related to a long prepuce, phimosis, scaling of the prepuce, poor hygiene, and high temperature, friction, and injury. Recently, it has been reported that infection with human papillomavirus (HPV) 16, 39, 8, or 51 plays a role in pathogenesis of these diseases [1].

2. Clinical Presentation

A 36-year-old man with erythema of the glans was admitted to our clinic in November 2011. Three years earlier, a mung bean-sized, infiltrating, pale-red patch appeared on the patient's glans, and was associated with slight swelling, some of which showed a few adhesive scales, without itching and pain. Therefore, the patient was not treated. The erythema gradually extended to the penis and prepuce, involving nearly two-thirds of the glans, penis, and prepuce, and showed significant infiltration. More recently, the glans erythema developed miliary-sized papules, which gradually increased to soybean-sized nodules. Previously, no circumcision had been performed, and a history of unclean sexual intercourse and drug allergy was denied. Family members had no similar medical history. HPV16 (+), HBsAg(-), Anti-HCV(-), Anti-HIV1+2(-), TPPA(-), RPR(-). Other routine examinations, such as blood, urine,

stools, biochemical tests, chest x-ray for cardiopulmonary abnormalities, liver, gallbladder, pancreas, spleen, kidney, bladder, prostate, bilateral inguinal lymph nodes, and B-ultrasonography, were basically normal.

3. Physical Examination

The patient's general condition was good, and no abnormalities were found in all systems. Regarding dermatologic condition, a long prepuce, and soybean-sized, isolated, white nodules were seen on the glans, with a velvet-like appearance on the surface. Irregular patches (1.5 x 6 cm) were seen around the nodules and prepuce, with alternating red and brown color, and a slightly raised, clear border. Adhesive scales were seen, with an infiltration-like feel to touch (Figure 1a, Figure 1b). Bilateral inguinal lymph nodes were not significantly enlarged. Biosies of the glans were taken, and erythema and brown patches on the foreskin were sent for pathologic examination.



Figure 1a. The prepuce of the penis and the glans can be seen with well-defined, glossy, pigmented, non-uniformly infiltrating reddish-brown patches. A velvety nodule the size of a soybean is seen at the glans erythema



Figure 1b. After the first operation, a 10.5 cm vertical proliferative surgical scar with well-defined and infiltrative margins can be seen on the glans, with a glossy reddish-brown patch with uneven pigmentation.

4. Pathologic Results

The following findings were evident: excessive and incomplete keratosis; mass-like and papillomatous hyperplasia; local hypertrophy; extrusion and extension to the dermis; cell proliferation; disordered cellular arrangement, with heteromorphic cells, vacuole cells, dyskeratotic cells, obvious nucleoli, and pathologic nucleolar division; and superficial dermal vasodilation with dense infiltration of lymphocytes, plasma cells, and neutrophils. These results were consistent with verrucous carcinoma (Figure 2, Figure 3a, Figure 3b). We observed excessive and incomplete keratosis, slight hypertrophy of the spinous layer, loss of polarity of spinous cells, disordered arrangement, multiple dyskeratotic cells, pathologic karyotype, obvious cell atypia, and lymphocyte and plasma cell infiltration around the superficial vascular layer of the dermis, which indicated EQ.

Figure 2, Figure 3a, Figure 3b. Epidermal hyperkeratosis, incomplete keratosis, hypertrophy of spinous layer, tumor mass in dermis, heteromorphic cells, vacuole cells, irregular nuclei, obvious nucleoli, and pathologic nucleoli division.

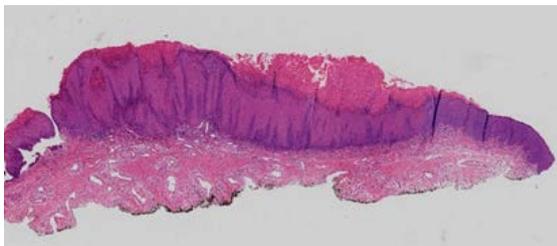


Figure 2. Verrucous carcinoma and Erythroplasia of Queyrat (HE x20). HE, hematoxylin and eosin

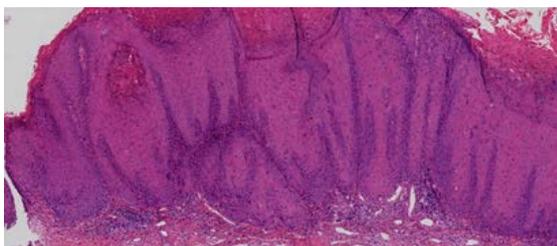


Figure 3a. Verrucous carcinoma (HE x50). HE, hematoxylin and eosin

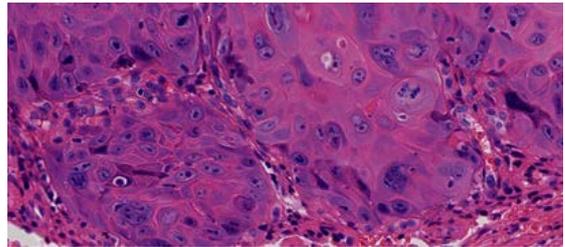


Figure 3b. Verrucous carcinoma (HE x400). HE, hematoxylin and eosin

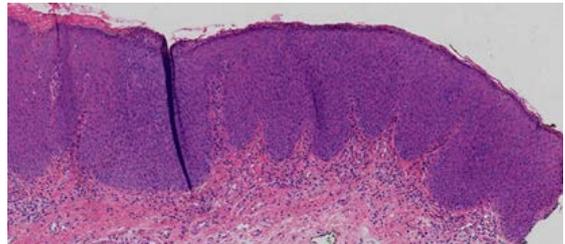


Figure 4. Erythroplasia of Queyrat (HE x100). HE, hematoxylin and eosin

Figure 2 and Figure 4. Mucosal epithelial hypertrophy, polar disorder of some heteromorphic cells, large nucleus, hyperchromatic cells, dyskeratosis, and lymphocyte and plasma cell infiltration around superficial vessels in the dermis.

5. Discussion

EQ, also known as erythematous hyperplasia, is a special type of squamous cell carcinoma in situ. It accounts of roughly 10% of all penile malignancies [2]. EQ can also occur in the urethra, vulva, oral mucosa, tongue, and conjunctiva [3]. Typical clinical manifestations are single or multiple well-defined, moist, velvety, glossy red plaques. Similar to Bowen's disease (BD), the presence of infiltration, nodules, and ulcers often suggests the possibility of transformation to invasive squamous cell carcinoma. EQ and BD are similar in histopathology, but location, pathogenesis, and biology of the tumors are different. EQ has a greater possibility of malignant transformation and metastasis than BD. EQ progresses to invasive squamous cell carcinoma in up to 33% of patients, approximately 20% of these patients will metastases [4], and prognosis is poor. In the present case, erythema first appeared on the glans, then gradually expanded to the penis and prepuce, with a few adhesive scales. Nodules, ulcers, and other malignant signs of invasive squamous cell carcinoma have not yet been observed.

Verrucous carcinoma is a very rare squamous cell carcinoma with good differentiation, slow growth, and low malignancy. It rarely metastasizes to local lymph nodes or distant areas [5]. However, it has strong invasiveness and a high recurrence rate. It was first described in 1896, It has also been reported in the oral cavity, anus, penis, and female genital apparatus [6]. The most common type is verrucous carcinoma of the penis, which accounts for 2.4~24.0% of penile cancers [7]. Phimosis and a redundant prepuce are two important factors causing verrucous carcinoma of the penis [8]. HPV infection is closely related to the incidence of penile

cancer and has been found in almost all penile cancer patients [9]. The present case occurred in the glans. The erythema showed a solitary nodule of soybean size, white, velvety shape, clear border, and HPV16 (+), which is consistent with previous reports mentioned above.

The clinical characteristics of verrucous carcinoma are very similar to those of condyloma acuminatum. Sometimes, verrucous carcinoma can be misdiagnosed as condyloma acuminatum because of superficial or inappropriate pathologic materials. Condyloma acuminatum occurs mainly in sexually active people, while most verrucous cancers occur in middle-aged or older people. Condyloma acuminatum occurs mainly in the lower genitourinary tract and perianal region, while verrucous carcinoma occurs mainly in the lower genital tract and skin. Invasive growth is a characteristic of verrucous cancer. Under the light microscope, the epithelial angle is widened and prolonged, and “pushed-forward” below the baseline of the stroma.

6. Treatment

Surgery is still the main treatment for EQ, including circumcision, simple lesion resection, Mohs operation, and partial or total penile resection. For some young patients, patients with large local lesions, or elderly patients who cannot tolerate surgery, local focus freezing, and drugs such as imiquimod cream, and 5-fluorouracil ointment, can be used for topical application. Carbon dioxide laser, and photodynamic therapy can be used before surgery to reduce lesions. Also, conservative treatment can be considered, and surgery can be delayed until after recurrence. Local excision is recommended for verrucous cancer, but inguinal lymph node dissection is not recommended. Radiotherapy is generally not recommended, and some scholars even forbid it, especially in patients with HPV infection. Some researchers believe that radiotherapy can induce interstitial changes in primary verrucous cancer and accelerate development of the disease [10].

For occupational reasons, our patient underwent only local excision of verrucous carcinoma of the glans. The skin lesion of EQ has not yet been treated and is still in follow-up.

7. Conclusion

EQ and verrucous carcinoma are rare squamous cell carcinomas *in situ* that may develop into invasive squamous cell carcinomas. In the clinic, we should detect and diagnose these conditions early, and initiate treatment early to minimize harm to patients. In addition, routine circumcision is an effective preventive measure.

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