



Anal Intraepithelial Neoplasia Associated with High-Grade Vulva Injury (Usual-Type Vulvar Intraepithelial Neoplasia): Surgical Treatment with Cutaneous Flap Advancement

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Abstract

The aim of the present article is to report the case of a young patient with bowenoid papulosis who was a carrier of other sexually-transmitted infections (STIs), such as HIV and high-grade vulva lesion (usual-type vulvar intraepithelial neoplasia, VIN), and to demonstrate the strategy used to manage the case, as well as to discuss important issues regarding the standardization of intraepithelial lesions.⁸

Keywords

- ▶ HPV
- ▶ anal neoplasia
- ▶ Bowen disease
- ▶ vulvar neoplasia

Introduction

The history of the terminology for lesions of the lower anogenital tract has taken two distinct directions, depending on whether the epithelial lesion is mucosal or cutaneous. The nomenclature of mucosal, cervical, vaginal, and anal lesions has been extensively developed by pathologists and gynecologists. And dermatologists and dermatopathologists have developed the nomenclature for cutaneous vulvar, penile and perianal lesions. The terms for HPV-associated lesions in the anogenital tract have changed numerous times in recent years, along with our understanding of the disease and the treatment strategies.

A study group was formed to unify the histopathological nomenclature of epithelial lesions caused by HPV in the

lower anogenital tract. And one of the recommendations is to use the appropriate intraepithelial neoplasia (IN) terminology without specifying the location. And when mentioning a specific location, the appropriate full term should be used. Thus, for a lesion in the cervix, cervical intraepithelial neoplasia (CIN), in the vagina, vaginal intraepithelial neoplasia (VaIN), in the vulva, vulvar intraepithelial neoplasia (VIN), in the anus, anal intraepithelial neoplasia (AIN), in the perianus, perianal intraepithelial neoplasia (PAIN), and, in the penis, penile intraepithelial neoplasia (PeIN).^{1,2}

Perianal intraepithelial neoplasia is a sexually transmitted infection (STI) caused by the human papillomavirus (HPV) which affects the skin of the anogenital region. The predominant symptom is pruritus, and pain or some type of local discomfort may also occur.³

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The diagnosis of bowenoid papulosis is clinical because its appearance and location are characteristic. The lesion is characterized by multiple small flat or velvety papules of a color ranging from pink to dark brown.

Together with Bowen disease (BD) and Queyrat erythroplasia, high-grade AIN, a precursive lesion of squamous cell carcinoma (SCC), is considered. Even without treatment, most lesions remain benign and stable, a fact that is more common in bowenoid papulosis. In BD, the lesions may have a worse prognosis, and are more susceptible to malignancy. Regarding treatment, various modalities are available, from topical to immunotherapeutic and ablative medications.⁴⁻⁷

The aim of the present article is to report the case of a young patient with other STIs, such as HIV and high-grade vulva lesion (usual-type VIN), and to demonstrate the strategy used to manage the case, as well as to discuss important issues regarding the standardization of intraepithelial lesions.⁸

Report

A 45-years-old female patient attended the coloproctology outpatient clinic complaining of itching and discomfort in the anus and perianal region. She reported being HIV-positive and having used antiretroviral therapy (ART) for 10 years, and had already undergone surgical treatment for usual-type VIN.

Upon examination, we identified involvement of the entire perianal region by an extensive lesion, of irregular edges, formed by multiple small papules, coalescing, with a color ranging from pink to dark brown. ► **Figure 1**.

Anal material was collected for cytology.

As an initial treatment, we opted to use 25% podophyllin. Weekly application was performed for five weeks, without success. We changed the treatment strategy to the use of imiquimod cream at 5%, 3 times a week, for 16 weeks. As there was no adequate clinical response, we decided to perform the excision of the lesion.

During surgery, we resected the area of skin that contained the entire lesion viewed macroscopically, and then, to correct the skin defect, we performed the reconstruction at the same surgical time with V-Y flap advancement (► **Figure 2**).

Subsequently, the patient was followed up at the coloproctology outpatient clinic and the dressing committee (► **Figures 3 and 4**).

The histopathological result revealed a high grade AIN, previously described as NIA 3 or carcinoma in situ.

Anal cytology revealed a low-grade AIN.

The patient remains under follow-up and semiannual outpatient observation.

Discussion

Bowenoid papulosis, along with BD and Queyrat erythroplasia, are considered high-grade IN, precursive lesions of SCC.

Bowenoid papulosis and BD are histologically indistinguishable, so the diagnosis becomes clinical. Bowen disease is most often asymptomatic, and its macroscopic aspect can vary greatly, from erythematous-desquamative plaques with well-defined contours to vegetative lesions. Another aspect that differentiates them is the fact that BD may have a worse prognosis and it is more susceptible to malignancy.

Among the pathologies to be considered in the differential diagnosis of bowenoid papulosis, we should mention, in addition to BD and Queyrat erythroplasia, Paget disease, basal cell carcinoma, seborrheic keratosis, chronic simple lichen, scleroatrophic lichen, acuminate condyloma, melanoma, eczema, descending carcinoma of the rectum, and inverse psoriasis.⁴⁻⁷

In the case herein described, we used two types of therapeutic modalities: topical medication for cytodestruction (podophyllin) and immunotherapeutics (imiquimod); however, only the ablative technique (surgical excision)



Fig. 1 Patient in jackknife position, lesion with features of bowenoid papulosis. In the first image, greater increase in the image of the vulva with aspect of previous surgery.



Fig. 2 Surgical procedure: resection followed by preparation of the V-Y flap advancement.



Fig. 3 On the 30th postoperative day, the closest image of the anus shows a partial dehiscence of the flap suture.



Fig. 4 Final aspect on the 120th postoperative day: in the first image, patient in lithotomy position, and in the following two images, in jackknife position. Healing with good appearance and epithelialization near the anus in the area that previously presented dehiscence.

succeeded. Therefore, the coloproctologist should be able to surgically remove the lesion when the clinical treatment fails.

Another fundamental issue in the management of this case, especially regarding the follow-up, is that the professionals involved (surgeons, pathologists, and oncologists) should be accustomed to the specific and unified terminologies of the lower genital tract. Therefore, in the present report, we used the Lower Anogenital Squamous Terminology (LAST) and the classification of the International Society for the Study of the Vulva and Vagina Diseases (ISSVD) to avoid conduction errors and unfavorable outcomes.^{2,8}

Unfortunately, a classic example of this error is to receive results of carcinoma in situ and treat this as an invading carcinoma. We must remember that, in carcinoma in situ, high-grade AIN, the lesion respects the limits of the basal membrane, and there is no invasion of the lamina itself, that is, there is integrity of the basal membrane of the epidermis. Therefore, initiatives such as LAST and ISSVD have emerged to standardize the diagnosis of HPV-induced squamous epithelial lesions of the lower anogenital tract.⁸

An increasingly common aspect of great relevance for the case herein reported is the association of HPV-induced lesions in both the gynecological region (vulva, vagina, and

uterine cervix) with anal lesions, even if the patient does not have anal sexual intercourse. We see this association with great frequency in the clinical practice and in recent studies, which may indicate that HPV infection is a multicentric disease.

As recurrences are frequent, these individuals should be followed up in specialized services indefinitely, to identify recurrence or progression of the lesion.

Conflict of Interests

The authors have no conflict of interests to declare.

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