

BUSCHKE — LOWENSTEIN ANAL TUMOR: AN AMBIGUOUS ENTITY

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The Buschke — Lowenstein tumor is a rare sexually transmitted disease. Its location at the anal margin is also very rare. The most incriminated risk factor is human papillomavirus infection. Its clinical form may be confusing with other tumor and infectious lesions. Histologically, it is characterized by a well-differentiated malpighian proliferation. It represents local aggressive behavior. The treatment of reference remains the surgery with healthy margins of excision. Other treatments have been tested, but their effectiveness remains uncertain. We report here a new case of anal margin Buschke — Lowenstein tumor with a review of the literature.

Key Words: Buschke — Lowenstein tumor, epidemiology, treatment.

The Buschke — Lowenstein tumor (BLT) is a rare tumor. It reaches 0.1% of the population in sexual activity [1, 2]. Its location at the anal margin is also very rare. It represents a sexually transmitted disease. The most incriminated risk factor is human papillomavirus (HPV) infection. Histologically, it is characterized by a well-differentiated malpighian proliferation. It represents local aggressive behavior. The treatment of reference remains the surgery with healthy margins of excision. We report here a new case of anal margin BLT with a review of the literature; we will try to identify the epidemiological, clinical, therapeutic and evolutionary aspects of this tumor.

Case report

We report the case of a 47-year-old patient. Informed consent was obtained from the patient. He was operated several times for recurrent abscesses of the anal margin. The anatomopathological examination concluded each time to a condyloma acuminata. The decision was then to make a large surgery to limit the risk of recurrence. Abdominoperineal amputation was performed (Fig. 1). Extemporaneous examination showed a verrucous carcinoma of giant condyloma or BLT of the anal margin extended to the anorectal wall, infiltrating all tunics with perforation (Fig. 2). The surgeons completed with lymph node dissection. Sixteen regional lymph nodes were negative. The surgical margins were healthy. The tumor was classified pT3N0 according to the TNM classification. Two months after surgery, the tumor recurred as an extensive infected budding lesion with cauliflower appearance (Fig. 3). Computed tomography showed a largely necrotic 16 cm perineal mass invading the internal obturator muscles, the precoccygeal region, the prostate and the bladder, with bilateral iliac lymph nodes, with no secondary lesions at a distance. The radiotherapy (RT) was judged technically

not feasible because of the large irradiation field. The decision was then to conduct chemotherapy with capecitabine (825 mg/m² × 2/day for 21 days) + cisplatin (100 mg/m² day 1), every 21 days.

DISCUSSION

BLT is a relatively rare tumor. It was first described in 1925 by Buschke and Lowenstein, who initially



Fig. 1. Abdominoperineal amputation specimen with a 6.5 × 3.5 cm polypoid white budding lesion

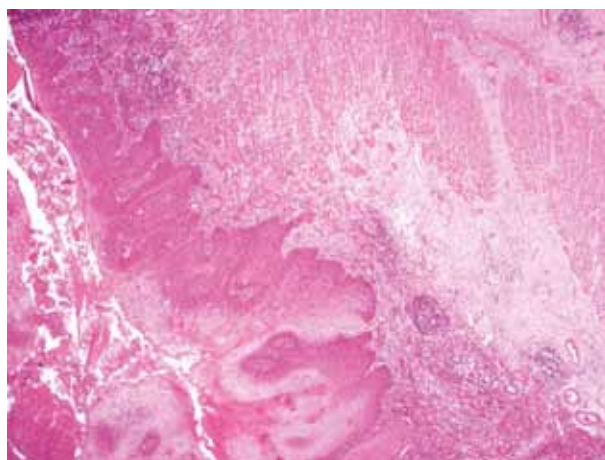


Fig. 2. Well-differentiated squamous proliferation with verucous papillomatous appearance

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Abbreviations used: BLT — Buschke — Lowenstein tumor; HPV — human papillomavirus; RT — radiotherapy.



Fig. 3. Infected budding lesion with cauliflower appearance

named it “condyloma acuminate carcinoma-like” [3]. It is a sexually transmitted disease [2]. It can reach both sexes, with a male predominance. It comes most frequently in men between the 4th and 6th decades, similar to our case [4]. HPV, and specifically serotypes 6 and 11, is incriminated in the genesis of these tumors [3, 5, 6]. Transformation of condyloma into BLT can be controlled by the immune system [7]. Thus, this tumor is frequently associated with congenital or acquired immunodepression (AIDS, immunosuppressive therapy, alcoholism, diabetes, chemotherapy). In our case, the 47-year-old patient was immunocompetent and had not reported the notion of heterosexual behavior.

The external genitalia are the preferred localization of this tumor. BLT is localized in the anorectal area in 10 to 17% [6]. Our patient was operated several times for recurrent abscesses of the anal margin, which represents the mean reason of consultation [3].

In our case, the lesion was preceded by the appearance of recurrent condyloma acuminate, which concords with data of literature [1, 6].

Clinically, the tumor appears as a rather bulky mass, budding, of cauliflower aspect, which evolves in surface and in depth. Differential diagnosis are represented by other tumor lesions (squamous cell epitheliomas) or infectious lesions (syphilis in its secondary form, verrucous and vegetative tuberculosis, Nicolas Favre’s disease, inguinal granuloma or donovanosis, anogenital amoebiasis) [8]. Associated sexually transmitted infection must be systematically sought. The tumor can grow into an enormous mass, exceeding tens of centimeters, repressing and even destroying adjacent structures and organs. It is thus known by its locoregional aggressiveness, as in our case. It is also characterized by a high relapse rate, reaching up to 66% after excision, according to the

literature [9]. On the other hand, metastases are exceptionally described [3, 10].

Histologically, the exact terminology of this tumor remains controversial. Some authors consider it a real squamous cell carcinoma. Others consider it to be a transitional form between condyloma acuminata and epidermoid carcinoma [3, 5]. Histologically, BLT is characterized by a well-differentiated squamous proliferation. It results in exo- and endophytic hyperpapillomatosis with hyperacanthosis. The hyperplastic epithelium is well differentiated, regular, without cytonuclear abnormalities. The basement membrane is respected, which is in favor of the benignity of the tumor despite its aggressive behavior [4, 6, 11]. The treatment of these tumors is not consensual due to its rarity. Several therapeutic modalities have been proposed. Surgery represents the gold standard of treatment [3, 12]. This must be as wide as possible [9], in order to reduce the risk of recidivism that exceeds 60%. However, given the large volume of the tumor, operability is not always accessible. Our patient was operated with healthy margin; however the tumor recurred in two months, as an infected mass. Thus the use of other treatment modalities is sometimes necessary. Several types of treatments including topical chemotherapy with fluorouracil, laser excision and tumor destruction by cryotherapy or electrocautery, have been described and tested in reported cases from the old literature, and found to be enough active and potentially effective [3, 13, 14]. Intralesional or local application of interferon has also demonstrated limited efficacy [3]. RT has been tested and has contributed to satisfactory results. However, some are wary of its indication given the risk of degeneration into anaplastic carcinoma that it could induce [12, 15]. Chemotherapy alone or concomitantly with RT was also reported in some series and reported cases. It was most often based on 5-fluorouracil and cisplatin. This type of protocol has been shown to be effective with satisfactory response and tumor volume reduction rates. Other protocols based on methotrexate have also been reported in the literature [12]. Treatment with concomitant radiochemotherapy may be the exclusive curative treatment, or represent a neoadjuvant treatment to achieve and facilitate subsequent surgery. Our patient was proposed to have chemotherapy (capecitabine and cisplatin) to allow secondary local RT. In fact RT was impossible due to the large irradiation field. Despite the combination of several therapies, the prognosis of this tumor remains poor, given the risk of recurrence, locoregional aggressiveness, infection, degeneration into a “frankly” malignant tumor, and mortality exceeding 20% [9, 12]. Larger series and randomized studies could solve the question about the optimal treatment of these rare tumors.

CONCLUSION

Anal margin BLT is a rare tumor. It is characterized by its local aggressiveness but also by a low metastatic potential. Some authors consider it as a real squamous

cell carcinoma. For other authors, it presents a benign tumor and a form of transition between a condyloma acuminata and squamous cell carcinoma. Therapeutic management is not yet univocal. Large surgery remains the gold standard, but sometimes it is not accessible given the large tumor size often observed. Concomitant radiochemotherapy, reported in several cases in the literature, gives satisfactory results with good responses and significant reduction in tumor size. The optimal therapeutic strategy remains to be defined by other studies of larger series.

CONFLICT OF INTEREST

None.

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