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Giant condyloma acuminatum of the anorectum: successful radical surgery with anal reconstruction

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ABSTRACT

Buschke-Löwenstein tumor, or giant condyloma acuminatum, is a relatively uncommon lesion of the anus with aggressive local invasive behavior which may present as a large warty tumor of the genital region with expansive and destructive growth. Many sporadic reports have been published suggesting various therapeutic strategies. We report a case of Buschke-Löwenstein tumor treated with conservative surgery followed by reconstructive procedures without a loop colostomy.

Introduction

Buschke and Löwenstein described in 1925 a slow-growing, locally aggressive, destructive and disfiguring cauliflower-like tumor that typically affects the anogenital and perianal regions as the "giant condyloma of Buschke and Löwenstein" (GCBL)[1]. Along with oral florid papillomatosis and epithelioma cuniculatum, GCBL is considered a regional variant of verrucous carcinoma.

GCBL is a rare variety of squamous cell carcinoma, a low-grade wart-like tumor believed to be caused by human papillomavirus (HPV) infection. In some cases it presents as a large warty tumor of the genital region with expansive and destructive growth.

Many sporadic reports have been published suggesting various surgical and/or nonsurgical therapeutic strategies. However, due to the nature of the disease, no definite treatment modalities have been established. GCBL lacks basement membrane invasion and is therefore, according to histological criteria, not a malignant lesion; nevertheless, it often shows areas of cellular atypia where invasion is noted, characterizing it as a putative precancerous lesion[2]. Furthermore, it has a significantly high recurrence rate after surgery (44% in men and 18% in women)[3]. We here describe a case of GCBL of the perianal region that was successfully treated with conservative surgery and plastic reconstruction; the intervention did not include a loop colostomy.

Case report

A 54-year-old man was admitted to our hospital in February 2001 with a growth in the perianal region. On admission, the patient was affected by 2 concomitant tumors: a metastatic squamous cell carcinoma of the lung that was being treated with intravenous cisplatinum and fluorouracil, and a laryngeal squamous cell carcinoma for which the patient was receiving radiotherapy. The symptoms described by the patient were pain in the perianal region and difficulty of defecation.

At physical examination a large, ulcerated mass of 18x8x5 cm was observed in the perianal region (Figure 1). It was decided to remove it by wide surgical excision of the perianal region with a sphincter-saving procedure (Figure 2). Following demolition

Key words: giant condyloma, Buschke and Löwenstein, conservative surgery, sphincter-saving procedure.

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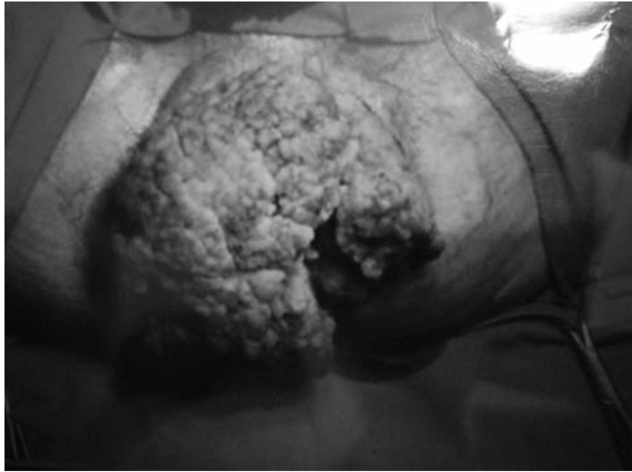

Figure 1 - Large ulcerated mass in the perianal region at physical examination.

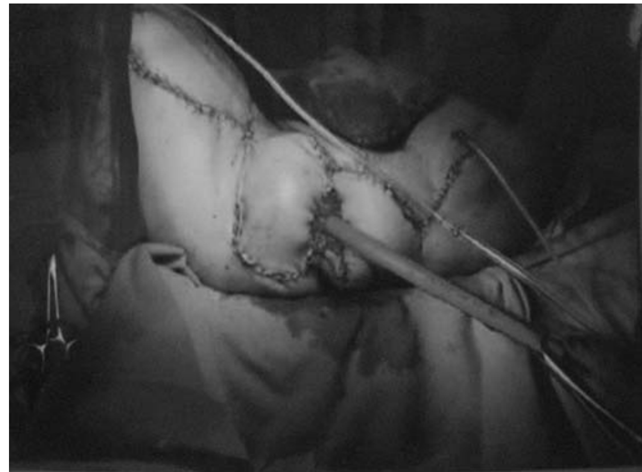

Figure 3 - Plastic reconstruction.

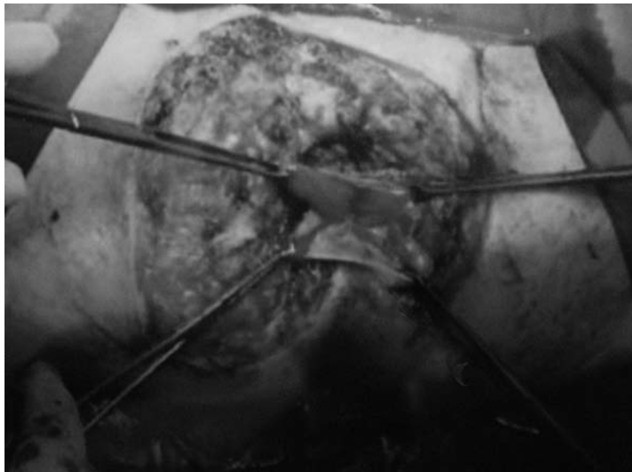

Figure 2 - Surgical excision with sphincter-saving procedure.

we marked a line from the pubic tubercle to the distal insertion of the gracilis tendon, then we drew a longitudinal skin paddle overlying the previous line. We detached the distal tendon of the muscle and raised gracilis myocutaneous island flaps bilaterally. These were transposed with a 90° rotation covering the defect and the donor site was closed directly. The anterior border of the flaps was sutured to the anal mucosa, paying particular attention that the edges of the mucosa were adequately evaginated (Figure 3). No postoperative complications were observed and no radiotherappy or chemotherapy was necessary.

Histological examination of the anal and perianal part of the lesion was performed in the perianal component. The epidermis was lengthened in a saw-toothed shape and parts of the epidermis invaded the deep layer of the dermis on low-power magnification. Although the nuclei were not atypical in high-power fields, koilocytes were observed in the epidermis while no invasion of the mucosa was observed in the anal part despite dense epidermal proliferation. On immunohistochemistry, anti-HPV polyclonal antibody staining (Dako, Japan) was positive in the koilocyte nuclei. Based on these results, the lesion was diagnosed as a GCBL.

Six years after surgery, no local or distant recurrences were documented. The patient died in 2007 of metastatic squamous cell carcinoma of the larynx.

Discussion

GCBL is a rare and peculiar type of tumor. It is characterized by semimalignant slow-growing condylomatous proliferation and often exceeds 5 cm in diameter. There seems to be a continuum between the HPV-induced condyloma acuminatum, the giant form of this process, and verrucous carcinoma[4].

There is considerable controversy regarding the optimal treatment of GCBL. Different therapeutic approaches including topical chemotherapy, radiotherapy, immunotherapy and radical surgery have been reported in the literature[5-11], but no standard treatment has been defined. Chemoradiation remains the therapeutic mainstay for anal malignancies but is not routinely employed in the management of GCBL without squamous cell carcinoma transformation[6]. Local surgery with electrocautery or laser is generally preferred over abdominoperineal amputation according to Miles[8]. Miles' procedure should be reserved for extremely extensive lesions or multiple, extensive recurrences. Due to the rarity of the disease, it is difficult to collect a large cohort of cases to define a targeted treatment protocol. Treatment is therefore chosen on the basis of single reports and individual experience[7]. Most authors prefer local or extensive abdominoperineal ex-

cision of the rectum followed by reconstruction using a transpelvic myocutaneous rectus abdominis flap[12]. We believe that conservative surgery should be performed when possible, followed by a reconstructive procedure[13].

Most reports seem to be in favor of a surgical approach in the treatment of GCBL of the perianal and anorectal regions. Chemotherapy and radiation therapy should be used only in cases of disease recurrence because their effectiveness has not been fully documented. Surgical planning should take into account the size of the lesion, the degree of penetration into adjacent structures, and lymph node involvement. If the surgery leaves a large scar, a skin graft may be necessary. Regular follow-up is essential to guarantee that the patient is disease free. If a GCBL recurs, further surgical resection, fecal diversion, and abdominoperineal resection in combination with chemo-radiotherapy will be necessary.

References

1. Buschke A, Löwenstein L: Über carcinomähnliche condylomata acuminata des penis. Klin Wochenschr, 4: 1726-1728, 1925.
2. Wietfeldt ED, Thiele J: Malignancies of the anal margin and perianal skin. Clin Colon Rectal Surg, 22: 127-135, 2009.
3. Reichenbauch I, Koebele A, Foliguet B, Hatier M, Masutti JP, Landes P: A Buschke and Lowenstein tumor in a female patient. J Gynecol Obstet Biol Reprod (Paris), 24: 491-495, 1995.
4. Sarzo G, Del Mistro A, Finco C, Frayle-Salamanca H, Marino F, Franzetti M, Ferrara R, Mistrangelo M, Savastano S, Vecchiato M, Merigliano S: Extensive anal condylomatosis: prognosis in relation to viral and host factors. Colorectal Dis, 12: e128-134, 2010.
5. Gholam P, Enk A, Hartschuh W: Successful surgical management of giant condyloma acuminatum (Buschke-Löwenstein tumor) in the genitoanal region: a case report and evaluation of current therapies. Dermatology, 218: 56-59, 2009.
6. Tytherleigh MG, Birtle AJ, Cohen CE, Glynne-Jones R, Livingstone J, Gilbert J: Combined surgery and chemoradiation as a treatment for the Buschke-Löwenstein tumour. Surgeon, 4: 378-383, 2006.
7. Mistrangelo M, Mobiglia A, Cassoni P, Castellano I, Maass J, Martina MC, Bellò M, Mussa A: Verrucous carcinoma of the anus or Buschke-Lowenstein tumor of the anus: staging and treatment. Report of 3 cases. Suppl Tumori, 4: S29-30, 2005.
8. Renzi A, Brusciano L, Giordano P, Rossetti G, Izzo D, Del Genio A: Buschke-Löwenstein tumor. Successful treatment by surgical electrocautery excision alone: a case report. Chir Ital, 56: 297-300, 2004.
9. Haque W, Kelly E, Dhingra S, Carpenter LS: Successful treatment of recurrent Buschke-Lowenstein tumor by radiation therapy and chemotherapy. Int J Colorectal Dis, 25: 539-540, 2010.
10. Gupta S, Malhotra AK, Verma KK, Sharma VK: Intralesional immunotherapy with killed Mycobacterium w vaccine for the treatment of ano-genital warts: an open label pilot study. J Eur Acad Dermatol Venereol, 22: 1089-1093, 2008.
11. Eftaiha MS, Amshel AL, Shonberg IL, Batshon B: Giant and recurrent condyloma acuminatum: appraisal of immunotherapy. Dis Colon Rectum, 25: 136-138, 1982.
12. Dolanc R, Kocher T, Langer I, Marti WR, Pierer G, Harder F: Malignant transformation of perianal Buschke-Löwenstein tumor. Extensive abdominoperineal rectum excision and reconstruction with transpelvic myocutaneous rectus abdominis muscle flap. Chirurg, 73: 370-374, 2002.
13. Parise P, Sarzo G, Finco C, Marino F, Savastano S, Merigliano S: Giant condyloma acuminatum of the anorectum (Buschke-Lowenstein tumour): a case report of conservative surgery. Chir Ital, 56: 157-161, 2004.