

Case Report

Giant Condyloma Acuminatum of the Ano-Genital Area: A Case Report and Current Trends in Management

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Abstract

Condyloma Acuminata is the common disease with cutaneous lesions, slow-growing, large, located furthermore in the ano-genital region. The incidence has sharply increased, with an estimated 2 to 3 million newly reported cases each year. Risk factors include anoreceptive intercourse, HIV and immunosuppression. Giant Condyloma Acuminatum (GCA) is a rare, rapidly growing, cauliflower-like tumor variant of condyloma acuminatum originally described in 1896 by Buschke and Lowenstein in 1925.

There is no general agreement on the choice of treatment for this tumor. Its management is often challenging due to the size, degree of local invasion and recurrence rate.

Wide radical excision with plastic reconstruction of skin defects seems to be the best treatment, while adjuvant therapies, such as radiotherapy, chemotherapy and immunotherapy, may achieve good results, but their effectiveness is still uncertain.

We report a case of GCA successfully treated by surgical excision alone and healing by secondary intention.

Keywords

Buschke-Lowenstein tumor; Condyloma acuminatum;
Surgical excision

Introduction

Giant Condyloma Acuminatum (GCA) is a rare disease that typically appears in the ano-genital region and presents rapidly-growing, large, cauliflower-like tumor, correlated to Human Papilloma Virus (HPV) infection. It was originally described as a penile lesion by Buschke in 1896 and Lowenstein in 1925 [1,2], while the first description of ano-rectal localization was by Dawson et al., in 1965 [3]. GCA tending to present in the fifth decade with a 2.7:1 male:female ratio. For

patients younger than 50 years of age, this ratio is increased to 3.5:1 [4,5].

The mean age at presentation is 43.9 years; 42.9 in males and 46.6 in females. There seems to be a trend toward younger age at presentation [6]. The most common presenting symptoms are peri-anal mass, pain, abscess or fistula and bleeding. Risk factors for disease contraction include ano-receptive intercourse diffused especially in the homosexual world, HIV and immunosuppression. Controversy exists as to the epidemiology, pathological nature and management of the tumor. Although clinically malignant, its histology is benign, without propensity for distant metastases. Despite this, GCA has a high rate of local recurrence with occasionally malignant

transformation to squamous cell carcinoma [7-9]. A lot of authors therefore consider GCA to be a regional variant of verrucous carcinoma [10,11] while others consider it a distinct and separate entity. There is no general agreement on the choice of the treatment for this tumor, due to its localization close to important structures (such as the anal sphincter) and to its biologic behaviour which is still not completely known.

Historically, treatment strategies have included topical chemotherapy, wide local excision, abdomino-pelvic resection, and the frequent addition of adjuvant and neoadjuvant systemic chemotherapy and radiation therapy. The authors report a clinical case of ano-genital GCA of a young woman 25 years old treated, with good outcome, by radical local excision, cauterization of the bottom and spontaneous complete wounds closure in second time.

Case Report

A 25 year old woman suffering from serious clinical form of Systemic Lupus Erythematosus (SLE) with haematic, renal and pulmonary involvement. Beginning of disease at the age of four with haemolytic autoimmune anaemia and platelets reduction (Fisher-Evans Syndrome). She had immunosuppression with serious clinical form of nephritis treated with cortisone (MPR) and immunosuppressors (MFM, CICLOFOSFAMIDE, AZATIOPRINA) and positive to six clinical and laboratory symptoms of ARA classification. She was admitted to our Department in January 2014 for CGA located at the peri-anal and genital region, with progressive size increase and ulceration.

She referred normal sexual intercourses with a partner without skin or mucous lesions with condom usage. She didn't have HIV. Symptoms were pain in the anal region, bleeding, inability sitting and walking. At the physical examination this vegetative lesions presented as cauliflower-like, involving ano-genital area from coccyx tip to vulva on both sides and, laterally beyond the ischiatic tuberosity, foul smell.

The patient showed dysphonia.

The lesion was treated with local surgical excision and cauterization, performed in three different times. Wounds closure obtained by local medications with chlorexidina solution and antibiotic ointment. We performed three surgical times because of the extent of involving areas and risk of haemorrhage (after first surgical time the patient underwent blood transfusion).

The patient underwent an examination under general anaesthesia to explore involvement of the anal canal, rectum and vagina. The vaginal exploration showed the presence of a lot small lesions involving vaginal canal without uterine cavity. Several small condylomas were noted in the anal canal and rectum and as satellites around the main lesion. The endoscopy of the upper airway exploration was negative for lesion presence. During the second surgical procedure we

showed and performed excision of a smaller lesion on the mucous surface of lower lip with histological findings of squamous cellular papilloma. The post-operative period was early without complications. The patient continued to assume her immunosuppressive therapy and she went out of the hospital four days after operation with local medications. The pathologic analysis of the specimen confirmed the diagnosis of condyloma acuminatum, with moderate degree of dysplasia of epithelium, with koilocytosis atypia, acanthosis and parakeratosis. Human Papilloma Virus (HPV) analysis was performed on patient's tumor by Southern blot hybridization and revealed the presence of HPV types 6,11,16;18;31;3;35,45,51,52. After 2 years the patient is doing well with evidence of lower disease recurrence about small vaginal lips and anal sphincter controlled with local application of Imiquimod cream (Figure 1).



Figure 1: Left: Local findings before surgery; Centre: Local conditions after 3th surgical step; Right: Final aspect after wide local excision.

Discussion

It can still be very difficult to distinguish GCA from ordinary condylomas or squamous cell carcinomas. Clinically these three lesions present similar findings and can only be differentiated by histological examination [12]. On histology the main findings are acanthosis, thicker stratum corneum, marked papillary proliferation, focal parakeratosis, tendency to deep invasion, with displacement of the surrounding tissues [8,9]. These same features are seen also in verrucous carcinomas but, on the whole, GCA does not present histological evidence of malignancy, such as infiltration of basement membrane, lymphatic invasion, angio-invasion or distant metastases [13,14]. However squamous cell carcinoma or verrucous carcinoma can coexist with GCA in up to 50% of patients [6]. There is probably a viral aetiology (HPV subtypes 6, 11, 16, 18, 31, 33) [15,16].

Controversies exist about treatment that depends on lesions size and on local invasion degree. There are still many problems concerning the management of Buschke-Lowenstein tumors because of the high recurrence rates (67-70%), mortality percentage equal to 20-30 % and the lack of adequate series of patients following the same procedure [6,17,18].

Medical therapy avails as of use of Podophyllin, 5-Phluorouracile, Interferon, Imiquimod, Radiation therapy and Immunotherapy.

The topical application of resin of Podophyllin and also 5-Phluorouracile, even if gives good results on ordinary

condylomas acuminatum is not recommended in GCA for the lack of results [19].

Controversies exist about the role of radiation therapy because of evidence of anaplastic transformation, presence of new diffuses condylomas after its application and for poor follow-up [8,20,21]. Interferon injection directly in the lesion is safe and has an eradication rate of 47% to 62% but associated with high cost and a recurrence rate up to 40% [22,23]. Systemic Interferon may be considered for those lesions too large to be injected or excised surgically or in conjunction with other treatment to lower the recurrence rate. It is expensive, however, with a high incidence of side effects and a variable response rate [24]. The role of immunotherapy with preparation of autologous vaccine, has been evaluated in the treatment of recurrent extensive GCA, with very good results in small series of patients [25,26]. Good results are obtaining, recently, with Imiquimod, which is immunomodulator, used via topical as adjuvant therapy after surgical treatment for local recurrence control [27,28]. Surgical excision, when technically feasible, remains the mainstay of treatment [6,29,30]. Besides conservative surgical procedure (Mohs surgery; Liquid Azotum therapy, Laser therapy), useful for excision of small few and superficial lesions, exist two recommended surgical techniques:

- a) Wide surgical local excision with heal by secondary intention or reconstruction by skin grafts, local or distant pedicled flaps, free flaps [30,31].
- b) Abdomino-perineal resection, performed as last choice in cases of recurrence, pelvic invasion or malignant transformation [32].

Another problem about surgical treatment of GCA is tumor site. Many authors recommend performing temporary loop colostomy before surgery, to avoid the risk of fecal contamination of the wounds [3,14,18,30,32]. In our clinical case we have performed wide local excision, electrocautery and natural closure by secondary intention with local medications based on chlorexidina solution and antibiotic ointment. We used local applications of Imiquimod as adjuvant therapy in local recurrence control of disease.

Conclusion

GCA is a challenging problem for the reconstructive surgeon even today. A lot of techniques for GCA and condyloma treatment are described in the literature, but an ideal approach has not yet been found because of the lack of a consistent series of patients and the big changeability of clinical features expression. Despite this, after correct diagnosis, wide radical excision, when feasible, and plastic reconstruction of skin defects seems to be the best choice. Adjuvant therapies, such as chemotherapy, radiotherapy and immunotherapy, may achieve good results but their effectiveness is still uncertain.

References

1. Buschke A (1986) *Neisser's Stereoscopic Atlas*. Fischer, New York, USA.
2. Buschke A, Lowenstein L (1925) Über carcinomähnliche condylomata acuminata des penis. *Klin Wochenschr* 4: 1726-1728.
3. Dawson DF, Duckworth JK, Bernhardt H, Young JM (1965) Giant Condyloma and Verrucous Carcinoma of the Genital Area. *Arch Pathol* 79: 225-231.
4. Centers for Disease Control (CDC) (1983) Condyloma acuminatum - United States, 1966-1981. *MMWR Morb Mortal Wkly Rep* 32: 306-308.
5. Shulman S, Phair J, Sommers H (1992) *The biological and Clinical Basis for Infectious Disease*. (4th edn), Saunders, Philadelphia, USA.
6. Trombetta LJ, Place RJ (2001) Giant condyloma acuminatum of the anorectum: trends in epidemiology and management: report of a case and review of the literature. *Dis Colon Rectum* 44: 1878-1886.
7. Bogomoletz WV, Potet F, Molas G (1985) Condyloma acuminatum, giant condyloma acuminatum (Buschke-Lowenstein tumor) and verrucous squamous carcinoma of the perianal and anorectal region: a continuous precancerous spectrum? *Histopathology* 9: 1155-1169.
8. Creasman C, Haas PA, Fox TA Jr, Marta Balazs (1989) Malignant transformation of anorectal giant condyloma acuminatum (Buschke-Lowenstein tumor). *Diseases of the Colon & Rectum* 32: 481-487.
9. Chao MW, Gibbs P (2005) Squamous cell carcinoma arising in a giant condyloma acuminatum (Buschke-Lowenstein tumour). *Asian J Surg* 28: 238-240.
10. Batta AG, Engen DE, Reiman HM, Winkelmann RK (1990) Intravesical condyloma acuminatum with progression to verrucous carcinoma. *Urology* 36: 457-464.
11. Dianzani C, Bucci M, Pierangeli A, Calvieri S, Degener AM (1998) Association of human papillomavirus type 11 with carcinoma of the penis. *Urology* 51: 1046-1048.
12. Chu QD, Vezeridis MP, Libbey NP, Wanebo HJ (1994) Giant condyloma acuminatum (Buschke-Lowenstein tumor) of the anorectal and perianal regions. Analysis of 42 cases. *Dis Colon Rectum* 37: 950-957.
13. Alexander RM, Kaminsky DB (1979) Giant condyloma acuminatum (Buschke-Lowenstein tumor) of the anus: case report and review of the literature. *Dis Colon Rectum* 22: 561-565.
14. Renzi A, Giordano P, Renzi G, Landolfi V, Del Genio A, et al. (2006) Buschke-Lowenstein tumor successful treatment by surgical excision alone: a case report. *Surg Innov* 13: 69-72.
15. Boshart M, zur Hausen H (1986) Human papillomaviruses in Buschke-Löwenstein tumors: physical state of the DNA and identification of a tandem duplication in the noncoding region of a human papillomavirus 6 subtype. *J Virol* 58: 963-966.
16. Lehn H, Ernst TM, Sauer G (1984) Transcription of episomal papillomavirus DNA in human condylomata acuminata and Buschke-Löwenstein tumours. *J Gen Virol* 65: 2003-2010.
17. Rübben A, Beaudenon S, Favre M, Schmitz W, Spelten B, et al. (1992) Rearrangements of the upstream regulatory region of human papillomavirus type 6 can be found in both Buschke-Löwenstein tumours and in condylomata acuminata. *J Gen Virol* 73: 3147-3153.
18. De Toma G, Cavallaro G, Bitonti A, Polistena A, Onesti MG, et al. (2006) Surgical management of perianal giant condyloma acuminatum (Buschke-Löwenstein tumor). Report of three cases. *Eur Surg Res* 38: 418-422.
19. Krebs HB (1991) Treatment of genital condylomata with topical 5-fluorouracil. *Dermatol Clin* 9: 333-341.
20. Sobrado CW, Mester M, Nadalin W, Nahas SC, Bocchini SF, et al. (2000) Radiation-induced total regression of a highly recurrent giant perianal condyloma: report of case. *Dis Colon Rectum* 43: 257-260.
21. Bertram P, Treutner KH, Rübben A, Hauptmann S, Schumpelick V (1995) Invasive squamous-cell carcinoma in giant anorectal condyloma (Buschke-Löwenstein tumor). *Langenbecks Arch Chir* 380: 115-118.
22. Trofatter KF Jr (1991) Interferon treatment of anogenital human papillomavirus-related diseases. *Dermatol Clin* 9: 343-352.

23. Welander CE, Homesley HD, Smiles KA, Peets EA (1990) Intralesional interferon alfa-2b for the treatment of genital warts. *Am J Obstet Gynecol* 162: 348-354.
24. Browder JF, Araujo OE, Myer NA, Flowers FP (1992) The interferons and their use in condyloma acuminata. *Ann Pharmacother* 26: 42-45.
25. Wiltz OH, Torregrosa M, Wiltz O (1995) Autogenous vaccine: the best therapy for perianal condyloma acuminata? *Dis Colon Rectum* 38: 838-841.
26. Eftaiha MS, Amshel AL, Shonberg IL, Batshon B (1982) Giant and recurrent condyloma acuminatum: appraisal of immunotherapy. *Dis Colon Rectum* 25: 136-138.
27. Abcarian H, Smith D, Sharon N (1976) The immunotherapy of anal condyloma acuminatum. *Dis Colon Rectum* 19: 237-244.
28. Mestrović T, Cavčić J, Martinac P, Turčić J, Zupancić B, et al. (2003) Reconstruction of skin defects after radical excision of anorectal giant condyloma acuminatum: 6 cases. *J Eur Acad Dermatol Venereol* 17: 541-545.
29. Kraus SJ, Stone KM (1990) Management of genital infection caused by human papillomavirus. *Rev Infect Dis* 12: 620-632.
30. Oh C, Albanese C (1992) S-plasty for various anal lesions. *Am J Surg* 163: 606-608.
31. Uribe N, Millan M, Flores J, Asencio F, Díaz F, et al. (2004) Excision and V-Y plasty reconstruction for giant condyloma acuminatum. *Tech Coloproctol* 8: 99-101.
32. Dolanc R, Kocher T, Langer I, Marti WR, Pierer G, et al. (2002) [Malignant transformation of perianal Buschke-Lowenstein tumor. Extensive abdominoperineal rectum excision and reconstruction with transpelvic myocutaneous rectus abdominis muscle flap]. *Chirurg* 73: 370-374.