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Case Report

Saving Private Rights: An unusual case of genital warts
挽救私人權利：一個不尋常的性病疣個案

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A 72-year-old man presented with a 10-year history of phimosis. Circumcision revealed a large, warty mass on his glans. Biopsy demonstrated condyloma acuminata. Over one year they failed to respond to cautery, cryotherapy and topical imiquimod. Further biopsies confirmed that the lesion was histopathologically benign. Topical 5-fluorouracil (5-FU) was then commenced resulting in marked clinical improvement after two weeks, and apparent complete resolution at three months. One year later a small recurrence appeared and it again resolved with 5-FU. We report an unusual case of condyloma acuminata that highlights several important clinical, ethical and medico-legal considerations in the treatment and follow-up of such lesions.

一名七十二歲男子有著十年的包莖病史，包皮環切術後揭示了龜頭上有一大團疣狀腫塊，而活組織檢查確實為尖銳濕疣。經過一整年的治療，對燒灼、冷凍治療及外敷咪喹莫特等療法皆無效。重複活組織檢查確實其組織病理學上為良性，繼而開始外敷 5- 氟尿嘧啶治療；兩星期後腫塊得到顯著的臨床改善，三個月時更明顯地完全消失。一年後的細微復發，亦同樣被 5- 氟尿嘧啶有效消除。我們報告了一個不尋常的尖銳濕疣個案，以突出這問題的治療及跟進過程中，臨床、倫理及醫學法律各方面的重要考慮。

Keywords: 5-fluorouracil (5-FU), amputation, circumcision, condyloma acuminatum (CA), ethics, human papilloma virus (HPV)

關鍵詞：5- 氟尿嘧啶，截肢，包皮環切術，尖銳濕疣，倫理，人類乳頭瘤病毒

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Introduction

The Giant Condyloma of Buschke and Lownstein (GCBL) is a term that describes a bulky, locally destructive verrucous lesion typically appearing on the penis. It is characterised by large size and tendency to infiltrate deeper tissues despite a histopathologically benign appearance. It was first described by Abraham Buschke in 1886 and later

by Buschke and Ludwig Loewenstein in 1925.[1] Despite being an historical term, it is nonetheless still descriptively appropriate in certain cases such as the one we describe. However, having made this point, for the rest of this case report the clinical problem in question will be referred to as condyloma acuminata (CA).

Case report

An otherwise well 72-year-old man was referred to the dermatology clinic by his urologist for management of a warty tumour.

He initially presented to his primary care physician having been unable to retract his foreskin for over a decade, and was referred to urology because of phimosis. Circumcision was performed, revealing that the glans was covered with warts. A biopsy demonstrated CA with no significant cytological atypia and no evidence of malignancy (Figure 1) .

The patient reported that his wife had been his only sexual partner throughout his life. There was no prior history of warts in the couple and his wife had always had normal pap smears. The patient was screened for other sexually transmitted illnesses (STIs), and they were not detected.

The warts were cauterised but rapidly recurred. It was felt by urology at this point that a definitive surgical approach was preferred, but at the patient's request he was referred to dermatology to explore other management options.

On physical examination there were fairly large, verrucous lesions covering the entire glans penis and extending proximally to involve the remainder of the foreskin (Figure 2). The meatus was clinically uninvolved. There was no inguinal lymphadenopathy, and the scrotum and perineum were clinically disease free. A second biopsy was taken and it showed changes consistent with CA.

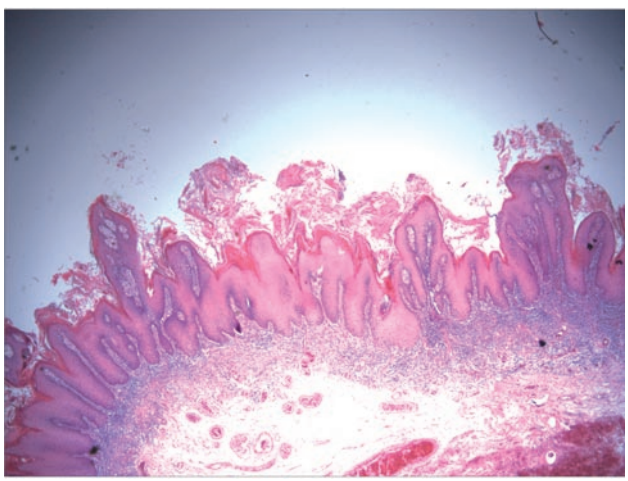

Figure 1. Histopathology prior to treatment. The section shows an exophytic and papillomatous proliferative lesion with features suggestive of condyloma acuminatum. There is no significant cytological atypia, and there is no evidence of malignancy. (H&E, original magnification x 5)

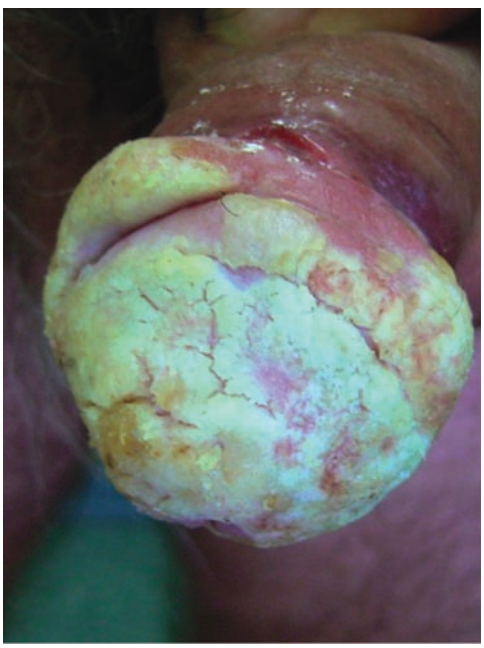

Figure 2. Hyperkeratotic, verrucous lesions covering the glans penis and distal foreskin.

About two months after first presenting to dermatology the patient was diagnosed with rheumatoid arthritis and was commenced on 5 mg of prednisone daily and 10 mg of methotrexate weekly.

The CA were initially debulked with salicylic acid ointment under plastic occlusion. Cryotherapy was then applied but the response was unimpressive. The patient was commenced on topical imiquimod. However, the improvement was slow, and a third biopsy was taken. This demonstrated a squamous proliferative lesion suggestive of CA, again with no evidence of malignancy.

After one year of treatment as outlined above, the patient was commenced on topical 5-fluorouracil (5-FU), since clinically bowenoid features surfaced. There was an excellent clinical response to this in just two weeks. After three months of 5-FU there was complete involution of the CA! (Figure 3) Towards the end of treatment with 5-FU, HPV genotyping for selected low and high-risk strains was performed on a biopsy, but the results were inconclusive. Accordingly, the need for longer-term observation and repeat relevant investigations was emphasized.

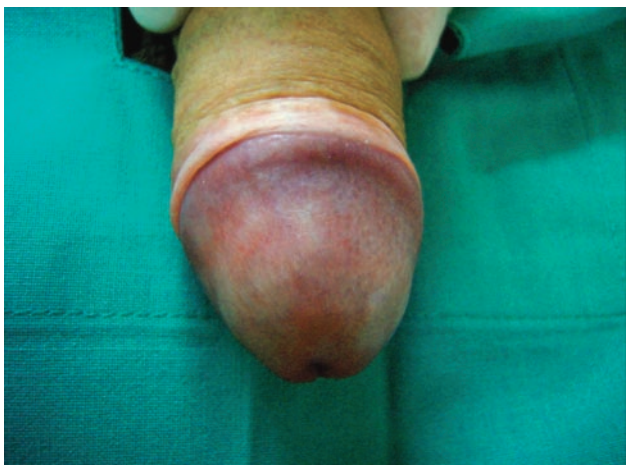

Figure 3. Three months following treatment with topical 5-fluorouracil.

The patient was reviewed at regular intervals and remained clinically disease free for a further twelve months. During this period he continued his treatment for rheumatoid arthritis. He then presented at a follow-up appointment with a small area of warts on his glans penis. At this point, he was advised to cease taking his immunosuppressant medications. The warts were again treated with a two-week course of topical 5-FU, again with complete resolution of the lesions. The patient remains clinically disease free at the time of this report, six months later.

Discussion

CA is a common STI, especially amongst young males. The prevalence is greatest in persons aged 17-33 years.[2] More than 70% of patients diagnosed with anogenital warts are aged 20 to 39 years, and beyond the age of 39 years, presentations for CA decrease as age increases.[3] There is an inverse association between age and HPV prevalence.[4] The most important risk factor identified for genital warts is the lifetime number of sexual partners.[5]

Not all cases of CA appear banal. Some, such as the one we report, evolve in a rather alarming manner. Our patient presented with CA relatively late in life. He was apparently immuno-competent and had no prior history of STIs. He only had one sexual partner. Despite this unremarkable history, he developed genital warts that proved to be recalcitrant and recurred swiftly after electrocautery, cryotherapy and topical imiquimod.

In view of the worrisome clinical developments, three penile biopsies were taken and no evidence of malignancy was detected. With the patient's agreement, and because of the emergence of clinically bowenoid changes, 5-FU was given a therapeutic trial. Almost surprisingly, there was a rapid involution of the CA, despite the context of

oral prednisone and methotrexate. It would appear that in selected cases when histologically proven benign CA fail therapies such as circumcision, cryotherapy, electrocautery and imiquimod, 5-FU should be considered, particularly if there is a history of immune-suppression.[6,7]

Our case also demonstrates the potential benefits of circumcision. There is strong evidence that circumcision reduces the risk of squamous cell carcinoma of the penis[8] and various STIs including HIV and syphilis.[9] There is also evidence that circumcision may lower the risk of HPV infection in males.[10] Furthermore, it simplifies topical treatment and enables easier examination of penile skin at follow-up visits. It is possible that circumcision could also be of benefit due to the removal of local irritants such as smegma and urine, which may cause penile pathology to progress.

Moreover, this clinical scenario highlights important medico-legal and ethical considerations. For instance, due to the alarming and rapid recurrence of the CA a surgical approach was initially preferred by the urologists. However, at the patient's request, options other than amputation were explored: amputation would have resulted in a poorer cosmetic and functional result, thereby decreasing quality of life. Had the amputation option been adopted, it could have resulted in profound psychological trauma. It is contentious whether such a drastic approach would have been in the best interest of the patient, given that biopsies repeatedly showed no evidence of malignancy. Furthermore, if the entire lesion was surgically removed and thorough histological examination demonstrated it to be benign, there could have been grave medico-legal implications.

The favourable outcome achieved in this case with topical chemotherapy stresses that if there are any doubts as to which treatment modality would be appropriate, the chosen approach should not be based on clinical appearances alone, and investigations such as multiple and serial biopsies should be performed if necessary. Most importantly, in clinical management dilemmas we must endeavor to preserve and respect private rights!

Finally, in cases such as this, regular and ongoing follow-up is of paramount importance, as demonstrated by the fact that the warts recurred after one year of apparent remission. The patient is clinically disease free at the time of this report. However, the responsibility of adequate follow-up remains with the treating team.

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