



A Giant Anorectal Condyloma Is Not Synonym of Malignancy

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Introduction

Anorectal giant condyloma acuminatum (GCA) or Buschke and Loewenstein tumor is a very rare sexually transmitted condition caused by Human Papillomavirus (HPV) infection which prevalence is around 0.1% among sexually active adults [1, 2]. GCA is a condition that has not been very well documented. All the literature we dispose of is based on case reviews and case reports. GCA was frequently described as a tumor because of its clinical aggressivity, contrasting with histological benignity [3]. It is also known for its high recurrence rate which is around 66% [4, 5] and its potential for malignant transformation (56%) [4]. The consequence of these features is the variability of treatment ranging from non-surgical treatment to mutilating surgery [6]. The purpose of this work is to present a case.

Case Presentation

A 30-year-old heterosexual male presented to our department with a large perianal tumor. We have no precise knowledge regarding the time span it took the lesion to reach its massive size. Perianal itch and discomfort when sitting were the only complaints the patient reported. A 15 pack-year smoking history was mentioned. No alcohol or drug use was noted. The patient denied any homosexual activity.

The physical examination revealed a 10 × 5-cm cauliflower-like mass, protruding from the intergluteal cleft seeming to cover completely the anus. On closer examination, the anal canal

looked free from invasion or other condylomata (Fig. 1). No adenopathies were palpable in the inguinal region.

The rectoscopy showed a healthy anal canal. A biopsy was performed to differentiate between GCA and a squamous cell carcinoma (SCC). It showed a papillomatous epidermis with multiple koilocytes and no atypia; however, the fragment did not include the dermis, so its invasion or lack of it could not be evaluated. This kept us from eliminating the SCC. Concerning the diagnosis of condyloma acuminatum (CA), the size of the tumor and the biopsy were not in favor of it.

Owing to the size of the tumor, complete surgical resection seemed to be the most effective treatment. After spinal anesthesia, the patient was placed in the lithotomy position. The whole perianal lesion was taken out in a single bloc (Figs. 2 and 3).

The histological examination of the excision revealed an exo-endophytic papillomatous epithelium with extensive hyperkeratosis and parakeratosis (Fig. 4a, b). It was largely composed of clear cells with numerous koilocytic changes (Fig. 4c). Neither atypia nor dermal invasion was noted. The



Fig. 1 Preoperative aspect of perianal cauliflower-like mass

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Fig. 2 Postoperative aspect of the perineum after excision of the tumor



Fig. 3 Macroscopic aspect of the resected specimen

excision was complete. The postoperative course was uneventful. There was no recurrence after a follow-up of 12 months.

Discussion

The main issue when dealing with GCA is to exclude the differential diagnoses and indicate the best treatment on which we have no clear consensus.

In our case, the definitive diagnosis of GCA could only be retained on the operative specimen. For this reason, the attitude of performing complete excision of GCA seemed to be the most appropriate initial treatment even in the absence of clinical signs of locoregional invasion. Additional treatment may be indicated based on the final pathological examination.

As in our case, the disease is most often seen in men in their 40s [5]. Indeed, the reported sex ratio is 3.2 male per 1 female and the average age of presentation is around 43.9 years [5]. The risk factors include anoreceptive intercourse, poor personal hygiene, immunosuppression, HIV, and chronic irritation such as perianal fistula or ulcerative colitis [4, 7]. The most common symptom in GCA is the perianal mass (47%) followed by pain (32%), abscess (32%), and bleeding [5]. Pruritus, difficulty in walking, and defecation have also been reported [4]. Our patient reported itching and discomfort especially in a sitting position but no pain.

The diagnosis of GCA is suggested by the clinical presentation and comforted by a biopsy. But the diagnosis of malignancy can only be excluded after a total excision and a histological examination of all the mass [6]. Areas of anal GCA and others of squamous transformation can be found within the same tumor [8]. GCA is indeed characterized by a high rate of malignant transformation ranging from 14 to 75% [5, 6]. The biopsy is therefore not sufficient to exclude malignancy and a non-surgical destructive technique such as laser vaporization is not recommended.

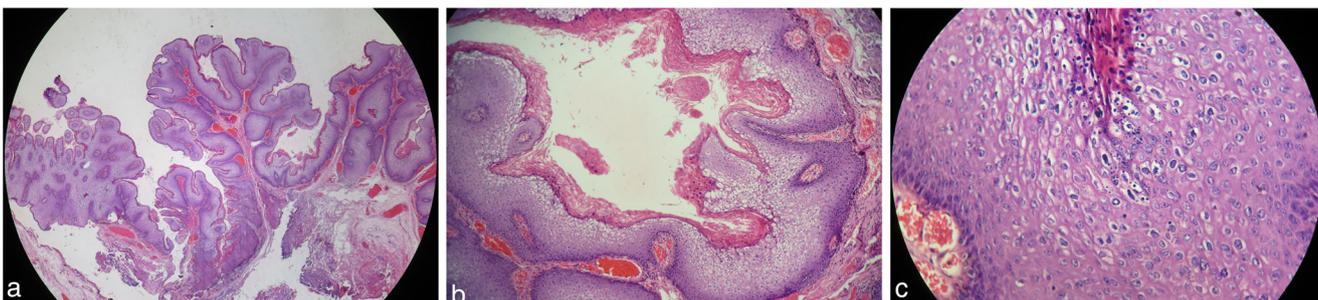


Fig. 4 Microscopic examination of Buschke-Lowenstern tumor (hematoxylin and eosin). **a** Hyperplastic, acanthotic papillomatous epithelium lining a normal dermis. **b** Extensive hyperkeratosis and

parakeratosis with pale cells. **c** Numerous koilocytes: big hyperchromatic nuclei within vacuolated keratinocytes

Clinical differential diagnosis of GCA, CA, and SCC is very difficult because all three have very similar presentations [4]. Many consider GCA to be an intermediate lesion between CA and SCC [8]. In fact, GCA is always preceded by CA and could transform into a SCC [2, 4]. GCA, known for its local aggressivity, is a slow growing, histologically benign circumanal mass, shaped in a cauliflower, with no infiltration of the underlying tissues [6, 9] which is not the case for SCC. Therefore, the diagnosis of squamous cell carcinoma cannot be eliminated before the postoperative pathologic test. It is also important to know that what differentiates CA and GCA is the presence of a very thick stratum, a pronounced papillary proliferations, and a tendency to grow in the latter [4, 9].

Clinical signs of invasion are suggestive of SCC and an MRI must be performed to gage the depth of the infiltration and guide the surgical decision [10]. In our patient's case, the MRI was not scheduled because the mass showed no signs of invasion. The tumor was limited to the perianal region with a healthy anus. The benignity of the tumor was later confirmed by the postoperative pathologic studies.

It is then safe to conclude that in the presence of a perianal mass with no signs of pelvic or nodal invasion and a positive biopsy for GCA, treatment could be started with complete resection of the tumor since it is very unlikely to be a SCC. The final histological examination will provide us with confirmation. In fact, surgery remains the treatment of choice regarding GCA with a success rate going from 63 to 91%, even after recurrence [7]; however, follow-up sessions are needed for an early diagnosis of a recurrence. If the biopsy reveals a SCC, authors recommend neoadjuvant chemoradiotherapy using the same scheme as in anal SCC [11], followed by abdominoperineal resection with negative tumor margins, then, depending on the case, reconstructive surgery. Abdominoperineal resection is also indicated in case of recurrences and pelvic invasion [4, 12].

Although non-surgical methods such as podophyllin injection have been reported as a treatment for GCA [4], they should, however, be avoided because they do not allow histological examination and can thus miss a malignant tumor.

Compliance with Ethical Standards

Conflict of Interest The authors declare that they have no conflict of interest.

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from all individual participants included in the study.

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