Giant condyloma acuminatum, initially described by Buschke and Loewenstein in 1925, is considered a verrucous carcinoma of the anogenital region associated with human papillomavirus (HPV). Clinically, these warts are characterized by their cauliflower-like appearance, their tendency to become infected and to form fistulas, and their capacity for local invasion. Despite aggressive therapy with multiple fulgurations, interferon alpha and isotretinoin, an invasive squamous cell carcinoma of the rectum developed. An abdominoperineal resection was done followed by radiotherapy and chemotherapy, but this treatment regimen was unsuccessful in controlling the progression of his carcinoma. Human papillomavirus (HPV) serotyping in tumoral tissue was positive for HPV types 11 and 16. In patients with giant anorectal condylomas associated with oncogenic HPV, the course of the disease may be aggressive, so they may benefit from early surgical and medical intervention.

**Case Report**

A 33-year-old heterosexual man was found to have anorectal condyloma acuminatum. The authors report on a 33-year-old man who was heterosexual and HIV negative and who had a giant anal condyloma. Despite aggressive therapy with multiple fulgurations, interferon alpha and isotretinoin, an invasive squamous cell carcinoma of the rectum developed. An abdominoperineal resection was done followed by radiotherapy and chemotherapy, but this treatment regimen was unsuccessful in controlling the progression of his carcinoma. Human papillomavirus (HPV) serotyping in tumoral tissue was positive for HPV types 11 and 16. In patients with giant anorectal condylomas associated with oncogenic HPV, the course of the disease may be aggressive, so they may benefit from early surgical and medical intervention.
immunologic work-up showed no abnormalities. There was rapid local recurrence, and treatment with interferon alpha (3 million units subcutaneously 3 times a week for 6 weeks) and then with isotretinoin (40 mg/d for 4 weeks) was unsuccessful.

Five months after the initial presentation, the patient presented with new perianal abscesses, multiple fistulas and condylomas extending 12 cm up the rectum. Histopathology of the rectal lesions showed a well-differentiated grade 1 squamous cell carcinoma infiltrating the rectal mucosa. The patient initially refused curative surgery, and only a derivation colostomy could be carried out for partial obstruction of the colon. Despite aggressive therapy to control local pain and infection, the patient’s condition worsened and an abdominoperineal resection was required (Fig. 2). For 4 months the patient’s condition was stable, then he had a recurrence of the carcinoma in the perineal scar. He received 42 Gy of radiotherapy to the pelvic area followed by chemotherapy with cisplatin and 5-fluorouracil. He then underwent laparotomy with resection of the ileum for intestinal obstruction and drainage of intra-abdominal abscesses. Microscopic examination of the resected ileum revealed the presence of condylomas on the mucosa (Fig. 3). There was no evidence of fistulas between the anal area and the ileum.

Three months later, the patient was admitted to hospital because of general deterioration. He refused any further treatment. He died 4 weeks later of complications of intestinal obstruction. At autopsy, a pelvic tumour measuring $7 \times 5 \times 5$ cm was found, with multiple carcinomatous lesions on the intestinal mucosa. There was no evidence of pulmonary or hepatic metastasis. HPV typing in tumoral tissue using polymerase chain reaction and molecular hybridization methods was positive for HPV types 11 and 16.

**DISCUSSION**

Over the last decade, the role of HPV in the development of certain genital carcinomas has been studied extensively. To date, 60 types of HPV have been identified. HPV types 16 and 18 have a high oncogenic potential and are typically associated with carcinomas of the penis and the anus. HPV types 6 and 11 are considered nononcogenic and are found in giant condylomas and in genital condylomas. The noteworthy finding of HPV 16 in our patient may explain the particularly aggressive nature of his disease. The association of HPV 16 with giant condyloma is consistent with the hypothesis that giant condylomas are part of the same spectrum of lesions as genital squamous cell carcinomas.

The finding of condylomatous lesions on the patient’s ileal mucosa without fistulas is also noteworthy. To the best of our knowledge this is the first such case in the literature. We suggest possible viral hematologic or lymphatic dissemination to explain this finding.

The contributing part of oncogenic HPV in malignant transformation remains uncertain. Other host factors or co-carcinogens may be implicated in the process of malignant transformation, such as immunosuppression, nutritional status or infection with HIV, cytomegalovirus or herpesvirus. Loning and associates have suggested that a decrease in patients’ cellular immunity may contribute to the development of genital carcinomas. It has indeed been shown that anal carcinomas are more frequent in AIDS and patients who have undergone renal transplantation. On investigation our patient had no immunologic abnormalities and no evidence of HIV infection.

Multiple treatments have been
used for giant anorectal condylomas. Podophylline, topical 5-fluorouracil, intralesional bleomycin and interferon alpha, isotretinoin, fulguration and systemic chemotherapy produce more or less satisfactory results. Since the natural history of giant anorectal condylomas is to malignant transformation, surgical excision with removal of the lymph nodes is currently the treatment of choice. Abdominoperineal resection is recommended in cases of multiple recurrences, invasion or frank malignant transformation. Our patient had multiple fulgurations and was treated with interferon alpha and isotretinoin, but this failed to control the progression of his condylomas. After a short delay in treatment because he refused surgery, an abdominoperineal resection was done followed by radiotherapy and chemotherapy. Despite these treatments, he died. Chu and associates suggested an aggressive course in patients with fistula filled with condyloma. Our patient had multiple condyloma-filled fistulas. HPV serotyping in tumoral tissue was only obtained after the patient’s death. Prior knowledge of the presence of an oncogenic HPV might have led to quicker surgical intervention followed rapidly by chemoradiotherapy, as suggested by Butler and colleagues. Early HPV serotyping of giant anorectal condylomas may, therefore, help identify patients who have associated oncogenic HPV. Those patients may have a rapidly deteriorating course of disease and may benefit from early, aggressive treatment.

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References


