Case Report

Squamous Cell Carcinoma Arising in a Giant Condyloma Acuminatum (Buschke-Lowenstein Tumour)

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Giant condyloma acuminatum (GCA) is a tumour that primarily affects the genital and perianal areas. Despite the histologically benign appearance, it behaves in a malignant fashion, destroying adjacent tissues, and is regarded as an entity intermediate between an ordinary condyloma acuminatum and squamous cell carcinoma. Primary anorectal lesions account for only a small number of GCA cases and, as with squamous cell carcinoma, the human papilloma virus is the causative agent. The hallmark of GCA is the high rate of local recurrence and transformation into squamous cell carcinoma. We describe a case of GCA complicated by malignant transformation, where locoregional control was achieved with combined chemoradiotherapy. [Asian J Surg 2005;28(3):238–40]

Key Words: Buschke-Lowenstein, chemoradiotherapy, giant condyloma acuminatum, human papilloma virus, squamous cell carcinoma

Introduction

Giant condyloma acuminatum (GCA) is a tumour primarily affecting the genital and perianal areas. It is also known as Buschke-Lowenstein tumour following the original description by these authors in 1925 of a lesion of the penis,1 the most common site for this tumour. Despite the histologically benign appearance, it behaves clinically in a malignant fashion, destroying adjacent tissues, and is regarded as an entity intermediate between an ordinary condyloma acuminatum and squamous cell carcinoma.2 Primary anorectal lesions account for only a small number of GCA cases and, as with squamous cell carcinoma, the human papilloma virus (HPV) has been identified as the causative agent.3–6 The hallmark of this disease is the high rate of local recurrence and transformation into squamous cell carcinoma.7 In this report, we describe a case of GCA complicated by malignant transformation, where locoregional control was achieved with combined chemoradiotherapy.

Case report

A 57-year-old heterosexual white male with a long history of severe psoriasis presented with a GCA with malignant transformation. His medical history was unremarkable apart from the psoriasis and he denied any risk factors for human immunodeficiency virus (HIV) infection. He had first noted an indurated lesion adjacent to his anus 1 year prior to diagnosis. Apart from mild perianal discomfort, there were no other symptoms. The treating physician made the clinical diagnosis of psoriasis and local therapy was delivered with topical steroids and, subsequently, imiquimod (Aldara™, 3M, Pymble, New South Wales, Australia). The lesion continued to grow, so a biopsy was performed. This revealed a giant anal condyloma containing well-differentiated squamous cell carcinoma with superficial invasion.

On examination, a large perianal tumour (8 × 6 cm in diameter) extended from the distal anal canal to the adjacent left buttock (Figure 1). No inguinal lymphadenopathy was
detected clinically or by computed tomography of the pelvis and inguinal region. No distant metastases were detected. Biochemical and serological investigations including HIV were normal.

The patient received primary combined chemoradiotherapy. Radiotherapy included 50.4 Gy to the primary tumour and prophylactic irradiation of the perirectal (45 Gy) and inguinal (36 Gy) lymph nodes. Chemotherapy consisted of mitomycin C at 10 mg/m² on day 1 and 5-fluorouracil (5-FU) at 1 g/m²/day delivered as a continuous infusion for 4 days in the first and fifth weeks of radiotherapy.

The patient had a clinical complete response to primary combined chemoradiotherapy, and had no evidence of recurrence at 12 months of follow-up (Figure 2). The patient denied any abnormal anal or bowel function.

Discussion

GCA is caused by HPV types 6, 11 and 16.³⁻⁶ Risk factors include immunosuppression, chronic irritation and poor personal hygiene.⁸⁻¹⁰ Chronic irritation secondary to perianal fistula and ulcerative colitis has been implicated in anal occurrence of this tumour.⁹,¹¹ The mean age of patients with perianal GCA is 43 years, with a male to female ratio of 2.2 to 1.⁹,¹¹,¹² The risk of recurrence after excision is 60–66%, with an overall mortality of 20–30%.⁹,¹¹,¹² Malignant transformation has been reported in 30–56% of cases.⁹,¹¹,¹²

The standard approach to perianal GCA is radical surgical excision. Patients with extensive lesions, especially those with multiple fistulous tracts and purulent discharge, may require a temporary loop colostomy. Any compromise regarding radicality is reported to predispose to local recurrence.⁹,¹³⁻¹⁵ For this reason, some have advocated abdominoperineal resection (APR) in cases with infiltration of sphincter muscles or the rectum.¹⁴,¹⁶,¹⁷

In patients with GCA with malignant transformation, the optimal treatment is not well defined. In a review of 11 patients treated with radical surgery, only five remained disease free.¹² This has prompted some to recommend preoperative chemoradiotherapy to downstage extensive tumours, followed by radical surgery.¹³ For classical anal canal cancers, the role of chemotherapy and radiotherapy is now well established and

Figure 1. A 57-year-old heterosexual male with squamous cell carcinoma arising in a giant condyloma acuminatum.

Figure 2. No evidence of locoregional recurrence 12 months after combined chemoradiotherapy.
the combined modality approach is superior to APR in terms of colostomy-free survival, with many patients maintaining good anal function.18

Several case reports have described patients with squamous cell cancers arising from GCA responding well to radiotherapy alone or chemoradiotherapy. Butler et al reported a patient with unresectable disease who received combined modality therapy.19 A pathological complete response was demonstrated following APR 32 weeks later. Chu et al treated a patient who had unresectable disease using preoperative chemoradiotherapy followed by APR and reconstructive surgery.11 This patient remained disease free at 22 months. Marsh et al reported a patient with extensive locoregional disease who was successfully palliated with chemoradiotherapy.20 Finally, Bertram et al described two patients with inoperable disease who proceeded to surgery following successful downstaging with moderate-dose radiotherapy.12

This case demonstrates the successful treatment of an invasive squamous cell carcinoma in the setting of GCA. A clinical complete response was demonstrated following chemoradiotherapy for a moderately large tumour. Further follow-up is required to determine whether this will be durable. This is consistent with the success achieved with large (T3–4) classical anal canal tumours, where 50–70% of patients attain a complete response. On the basis of this case and other published reports, chemoradiotherapy should be considered for patients with malignant transformation of GCA, with surgical salvage in those who fail to respond or subsequently progress.

References