Bowen's Disease of the Anal Canal Treated with Radiation Therapy: A Case Report

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Abstract:

Bowen's disease, also known as intraepithelial neoplasia, is a very slow-growing carcinoma in situ. It can progress to an invasive squamous cell cancer and infiltrate mucosa or skin in 3 to 5% of cases. The anal localization of this disease is very rare. Surgical resection is the standard of treatment. Radiotherapy keeps its place in recurrent or unresectable cases. We report a case of Bowen's anal canal disease in order to verify the effectiveness of Radiotherapy. He is a 59-years-old patient diagnosed with Bowen's anal canal disease at the National Institute of Oncology in Rabat. He was treated with exclusive radiotherapy at a dose of 60 Gray in 30fractions over 49 days. The evolution was marked by a complete clinical and radiological response and preservation of the anal sphincter with an 18 months follow-up.

Keywords: bowen's disease; anal canal; radiotherapy

Introduction:

Bowen's disease, first described by Johan Bowen in 1912[1], is high-risk intraepithelial neoplasia that appears primarily on the neck, face, genital organs and, perianal region. Anal localization is quite rare. The most important risk factor is infection with HPV 16 and 18[2, 3]. Biopsy with pathological examination provides the definitive diagnosis [4, 5]. The curative treatment is surgical excision with negative margins. Other less aggressive therapeutic methods can be discussed if the surgery is invasive with unacceptable morbidity or if the patient refuses, such as radiotherapy. We propose to evaluate the therapeutic efficacy of radiotherapy on this localized disease at the anal canal. We report the case of a patient diagnosed with Bowen's anal canal disease treated with exclusive radiotherapy.

Case presentation:

This is a 59-years-old patient with a non-significant medical history, followed for three years for recurrent anal margin condylomas after three surgical resections. The clinical examination revealed a tumor of the anal margin encompassing the anal orifice and at the rectal touch a rough appearance of the anterior wall of the anal canal (Figure1). The anoscopy showed a budding and friable lesion of the anal margin extended into the anal canal. The biopsy of the anal margin lesion shows a low-grade dysplasia on an acuminate condyloma with signs of HPV impregnation, while the biopsy of the anal canal lesion showed carcinoma in situ. The colonoscopy showed no other lesions.

The patient received exclusive 3D radiotherapy at a dose of 60 Gray in 30 fractions over 49 days. The treatment was well tolerated apart a Grade I radiation dermatitis treated symptomatically.

The evolution was marked by a good clinical and radiological response. A pelvic MRI performed at six months after completion of treatment concluded to a complete clinical response (Figure2). The follow-up consisted of a quarterly clinical examination and annual MRI that confirmed the absence of clinical or radiological recurrence with at 18 months follow-up.

Discussion:

Bowen's disease, also known as intraepithelial neoplasia, is carcinoma in situ. It is linked to several factors, including ionizing radiation, sun exposure, arsenic exposure [6], immunosuppression [7], human papillomavirus infection [8], genetic factors, trauma, chemical carcinogens and, X-rays. This disease can occur at any age, much higher in people aged 20-45 years old, with an equal incidence rate between the two sexes [9]. Bowen's disease of the anal canal is characterized by mild and infrequent clinical manifestations, such as burning or itching [10], with the appearance of perianal mass or bleeding in more than one-third of patients [11]. A therapeutic strategy was not recommended [12]. Resection was the preferred treatment. It is necessary to resect the entire lesion with negative margins to reduce the recurrence rate [13]. According to a study by Marchesa *et al*., the incidence of local recurrence of Bowen disease after surgical treatment depends on the surgical used technique. In the case of large excision surgery, this recurrence is at 23.1% with a margin greater than 1cm, 53.3% after local excision with a margin less

than 1 cm and, 80% after laser therapy [14]. It has been found that the wider was the surgery, the lower the recurrence was. But the surgical technique also depends on the tumor localization. In our case, performing large excision surgery and having a negative margin is uncertain because of the anatomical localization with the possibility of preserving the sphincter tonicity of the anal canal. Recently, several treatment modalities have been applied, including imiquimod [15], external fluorouracil [16], laser therapy, radiotherapy and, photodynamic therapy [17]. Of these, radiation therapy is appropriate for larger and recurrent lesions [12]. A study by Herat *et al*., had shown the efficacy of concomitant radio-chemotherapy in Bowen disease of the anal canal in an immunodeficient patient with good therapeutic tolerance and clinical response after 10 months of treatment [18]. Troicki *et al*., reported a recurrence of perianal Bowen disease initially treated with surgery. During this recurrence, the patient was treated with exclusive radiotherapy at 45Gy in 25 fractions with good tolerance and control at two years from the end of irradiation [19]. As for our patient, he is not immunodeficient, he was operated on three times for a condyloma of the anal canal. This time, Bowen's disease was diagnosed and he received exclusive radiation therapy of 60Gy in 30 fractions over 49 days. There was a marked clinical improvement and regression of the lesion during treatment. At 18 months of control, the patient had no visible lesion of the anal canal, and without palpable mass on the rectal touch. Sphincter preservation was also noted. Not only did this patient report a good quality of life with preservation of the anal sphincter but the most important is that the disease did not recur at 18 months of follow-up after irradiation. Radiotherapy is a therapeutic option in Bowen's anal canal disease that has shown its efficacy.

Figure1: Condyloma of the anal margin encompassing the anal orifice.

Figure2: Total clinical response in the anal canal and regression of the condyloma of the anal.

Conclusion:

Bowen's disease localized at the anal canal is a rare pathology. Different therapeutic methods have been evaluated. Radiotherapy is an efficient option that has proven to be effective with good local control.

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Disclosures

The authors report no conflict of interest concerning the case in this paper.

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