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Case Report

Squamous Cell Carcinoma On Buruli Ulcer Graft Scar, Ivory Coast

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Keywords

• Buruli ulcer; Scar; Squamous cell carcinoma

Abstract

Buruli ulcer is an infectious necrotizing panniculitis due to Mycobacterium ulcerans which heals leaving scars. On these scars, squamous cell carcinoma may occur in the long term, even in case of skin grafting. We report a case of squamous cell carcinoma occurring on a directed buruli ulcer scar.

Observation: A 35-year-old patient with a history of buruli ulcer healed with a skin graft in a specialized center for about 13 years, consulted for a cauliflower-like ulcerating swelling on the left elbow. The examination showed a large ulcerating cauliflower-like swelling. The diagnosis of squamous cell carcinoma was retained, and an amputation was performed without chemotherapy. There was no recurrence after six months of follow-up.

Conclusion: After a good healing, Buruli ulcer seems to present a risk of long-term evolution towards cancer. This observation raises the question of the carcinogenic role of mycobacterium ulcerans.

INTRODUCTION

Buruli ulcer is an infectious necrotizing panniculitis caused by Mycobacterium ulcerans [1]. Currently, the endemic continues to grow and its incidence is increasing dramatically, especially in West African countries such as Ivory Coast. Buruli ulcer is characterized by its chronic evolution, characterized by extensive skin eruptions complicated by dystrophic, fibrous and retractile scars [2,3,5]. On the other hand, directed healing gives considerable scars that are resistant to traumatic events. On these fibrous scars, squamous cell carcinoma may occur in the long term. In Abidjan, the first case was observed in 2010 [6], then eight cases were observed in 2015 [1] and finally one case in Bouaké in 2019. However, there is no on directed scars. We report a case of squamous cell carcinoma occurring on directed scarring of buruli ulcer in a 35-year-old patient without comorbidity.

OBSERVATION

A 35-year-old HIV-negative patient with a history of buruli ulcer healed with a skin graft in a specialized buruli ulcer management center in Kongouanou (Yamoussoukro) for about 13 years, consulted for an ulcerating swelling on the left elbow for the past 2 months. Examination showed a large swelling of about 12 cm in diameter, ulcerating and bubbling, with a cauliflower-like appearance, bleeding easily on contact, painful, and located on the inner side of the left elbow (Figure 1a,1b). The peri-lesional skin was normal in appearance. Biology revealed a normocytic hypochromic anemia. Histology revealed a proliferation of atypical squamous cells (large hyperchromatic nuclei, numerous mitoses) in invasive lobules, associated with disorders of keratinization. Finally, the tumor stroma is inflammatory (Figure 2). X-ray of the elbow showed bone lysis. The diagnosis of squamous cell carcinoma was retained without metastasis. Amputation was performed without chemotherapy. And there was no recurrence after six months of follow-up.

DISCUSSION

Buruli ulcer is hyperendemic in West and Central Africa : 16,517 cases were recorded from 2006 to 2015 and each year, about 500 new cases are recorded in Côte d'Ivoire, which remains a very active focus [4]. The epidemiology of the infection responsible for scarring [5,7] explains the young age of our patient and the location of the carcinoma on the limb. No comorbidity, in particular HIV infection, which is a factor favoring the development of malignant tumors in sub-Saharan Africa, was noted in our observation. Healing of the lesions occurred after several months of treatment. In our report, our patient had very good quality scars after directed healing. The occurrence of cancer in buruli ulcer scars is known [1,6,14].

Isolated cases of squamous cell carcinoma have already been described [8,9]. The first Ivorian observation was reported in 2010 [6]. Since then, eight cases have been recruited by the Abidjan center, which suggests a higher number at the national level since the Abidjan center does not have a monopoly on Buruli ulcer management. All these cases developed on fibrous and retractile scars. If sun exposure is the main risk factor for cutaneous squamous cell carcinoma in fair-skinned people, non-sun factors would be involved in people with pigmented skin. These are mainly chronic leg ulcers (neglected post-traumatic or

Figure 1 A and B: Burgeoning ulcer tumor.

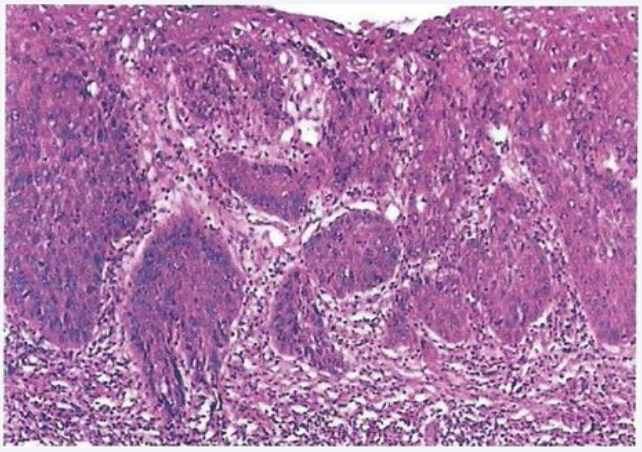

Figure 2 Histological appearance of squamous cell carcinoma.

infectious), HIV infection, discoid lupus and various chronic scars [10,11]. Carcinomatous degeneration of scars, including scars from old burns, is consistently reported.

There is a lack of epidemiologic studies on this topic in North African and sub-Saharan African countries where sunlight is high, medical resources are limited, and the risk of repeated scar ulceration becomes higher. The etiology of cancers occurring on scars is not fully understood, although current hypotheses include proliferation due to chronic inflammation and tissue irritation. In addition, ongoing tissue exposure to toxins and co-carcinogenic factors after injury, as well as poor vascularization of scar tissue, weakens local immune defenses [12-14]. The characteristics of Buruli ulcer scars, which resemble burn scars, may explain why they are particularly prone to carcinomatous degeneration. On the other hand, our patient benefited from directed healing and thus a better quality scar with good vascularization.

Moreover, one could also evoke the chronicity of the wound in this infection, or wonder if the mycobacterium itself could not play a role in carcinogenesis. This observation is, in our opinion, a warning signal. Given the number of people affected by this disease in their childhood or adolescence in Côte d'Ivoire and more generally in sub-Saharan Africa, it is to be feared that there will be a recrudescence of cases in the years to come when these adolescents reach adulthood. To this end, preventive measures should be taken from now on in the countries concerned: introduction of systematic surveillance of patients "cured" of Buruli ulcer in order to detect the first signs of carcinomatous degeneration and to sensitize the patients to an early consultation in front of any modification of their scars. This carcinological prevention requires very early management (at a stage without bone involvement or metastasis) of cases, could improve their prognosis.

CONCLUSION

After a complete healing, Buruli ulcer appears to have a long-term risk of progression to cancer. The scars of this condition, which could be considered as precancerous lesions. This observation calls into question the safety of directed healing or the carcinogenic role of mycobacterium ulcerans.

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