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A Giant Multifocal Squamous Cell Carcinoma Arising in a Patient with Hidradenitis Suppurativa

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Abbreviations: HS: hidradenitis suppurativa; SCC: squamous cell carcinoma; cSCC: cutaneous squamous cell carcinoma

1. Abstract

1.1. Introduction: Few reports describe squamous cell carcinoma arising in hidradenitis suppurativa. Although rare, the combination of SCC and HS is often associated with early metastasis and high mortality rates.

1.2. Case report: A 57-year-old man presented with a rapidly growing multifocal tumor in the gluteal area, which was affected by hidradenitis suppurativa. Physical examination revealed four exophytic, ulcerovegetative, erythematoviolaceous masses in his gluteal area with foul-smelling purulence on the surface. The biggest lesion was 10-cm long axis. He underwent a wide radical resection of the exophytic lesions. Histopathologic evaluation of the tumor showed 4 low grade well differentiated invasive SCCs with the following features: size varies from 1,5 to 11 cm, ulceration, and necrosis, with perineural or lymphovascular invasion. There was no distant metastasis on radiologic exploration. The patient died shortly after from septic complications. We would like to highlight the importance of prompt timely surgical excision with wide surgical margins.

1.3. Discussion: Squamous cell carcinoma arising in hidradenitis suppurativa is extremely rare with an overall incidence of 1%–3.2%. The predominant sites of cutaneous squamous cell carcinoma are the gluteal region, perianal region, and the genitals. It is characterized by a high mortality due to metastatic spread and sepsis. The management of cutaneous squamous celle carcinoma in hidradenitis suppurativa has not yet been standardized, but large surgical excision with at least 2 cm margins associated with radiotherapy appear to be the best therapeutic options. Considering the poor prognosis, prompt monitoring of hidradenitis lesions could help in preventing this dreaded complication.

2. Introduction

cSCC is a well-documented yet infrequent complication of HS. Its diagnosis could be particularly challenging because malignant transformation maybe easily mistaken for non-healing HS lesions. We hereby present a case of a 57-year-old man who presented with a rapidly growing multifocal tumor in the left gluteal area 30 years after the onset of HS with a fatal evolution.

3. Case Report

A 57-year-old man presented with a giant tumoral mass on his gluteal region that had developed in the previous 9 months. He had a 30-year history of HS grade 3, which was unsuccessfully managed with occasional oral and/or topical antibiotics and antiseptics. There was poor compliance since the patient was often absent from the follow-up visits. He had not used any biologic agents or undergone any surgery for HS. He had past history of smoking and cannabis use. A physical examination revealed four exophytic, ulcerovegetative, erythematoviolaceous masses in his gluteal area with foul-smelling purulence on the surface (Figure 1). The biggest lesion was 10-cm long axis (Figure 2). She also had abscesses; draining sinus tracts; and scarring on her axilla and groin regions. A wide radical resection of the tumor was performed by a general surgeon. Histopathologic evaluation of the tumor showed

4 low grade well differentiated invasive SCCs with the following features: size varies from 1,5 to 11 cm, ulceration, and necrosis, with perineural or lymphovascular invasion. Resection margins (periosteum) were tumorous. CT scan showed no distant metastasis. Radiotherapy was suggested after the surgery as an adjuvent treatment. Unfortunately, the patient died shortly after from septic complications.

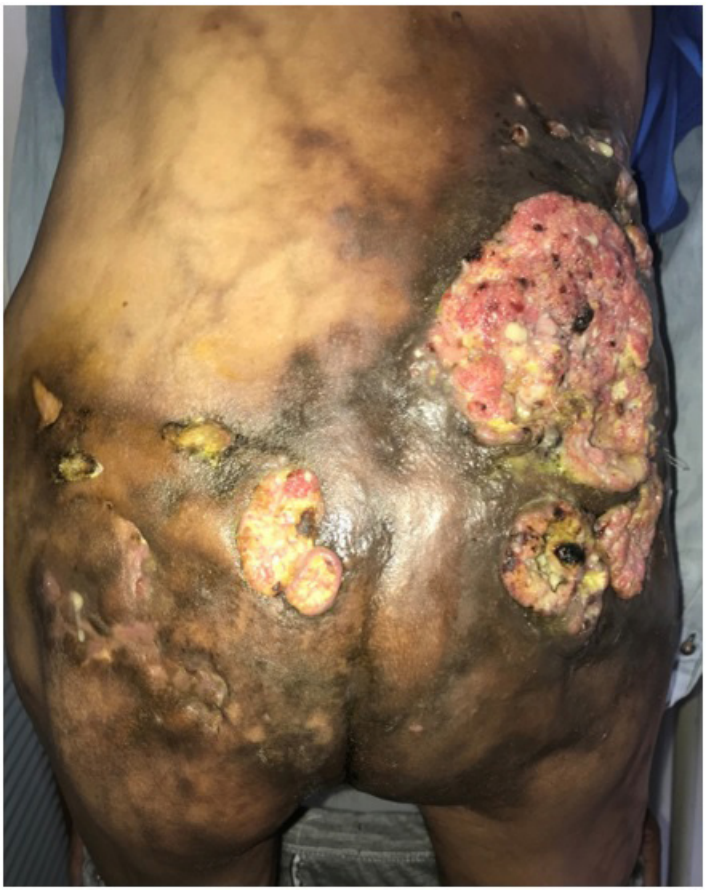

Figure 1: Exophytic ulcerovegetative erythematoviolaceous lesions in the gluteal area with foul-smelling purulence (posterior view)

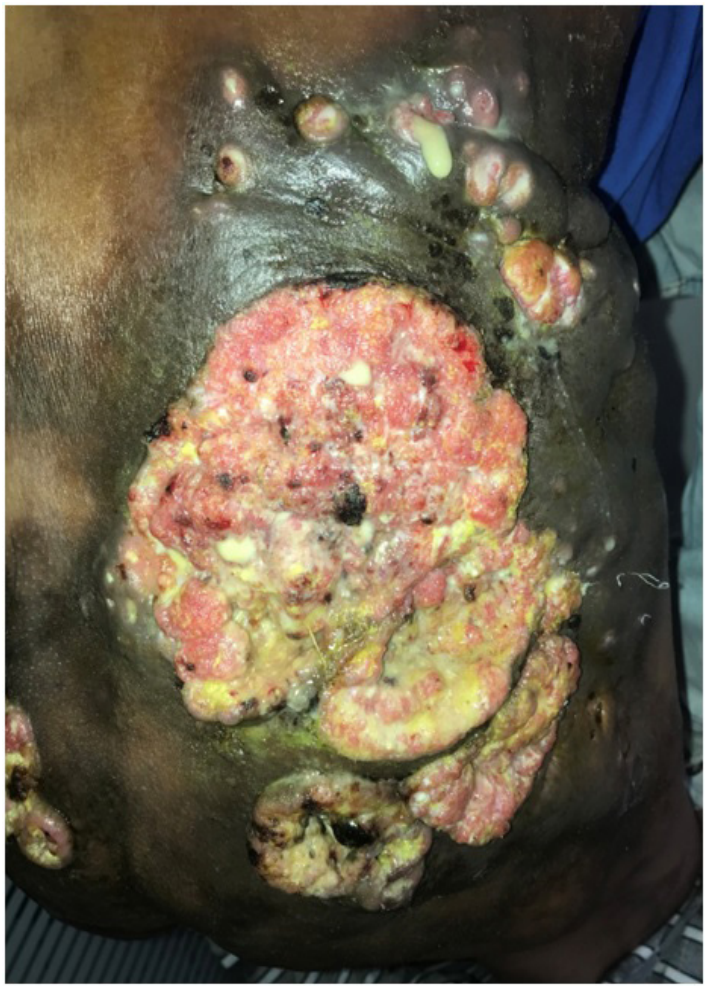

Figure 2: Largest exophytic ulcerovegetative lesion of 10-cm long axis (right gluteal area)

4. Discussion

HS was first described in the medical literature in 1839 by Alfred Velpeau [1]. It is defined as a chronic, inflammatory, recurrent, debilitating, skin follicular disease that usually presents after puberty with painful deep-seated, inflamed lesions in the apocrine-gland-bearing area of the body. The predominant sites of cSCC are the gluteal region, perianal region, and the genitals [2]. The average latency period from diagnosis of HS to the development of cSCC is of approximately 27 years, as was seen in our patient. With an overall incidence of 1%–3.2%, males appear to be at highest risk of developing this complication. A recent revue stated that human papillomavirus (HPV) and smoking might be risk factors for SCC in HS [3]. The precise mechanisms of pathogenesis of cSCC arising within HS lesions remain to be elucidated, yet two theories have been described in the literature. First, HS has been shown to be associated with inherited or acquired defects in the Notch signaling pathway. The latter plays a role in tumor suppression seen in non-melanoma skin cancers including cSCC, which could explain the association between cSCC and HS. Second, oncogenesis can be propagated by the chronic inflammation state of HS through dysregulation of tumor suppressor genes. The resulting immune response generates free radicals leading to oxidative damage implicated in malignant degeneration of longstanding HS to cSCC [4].

Mortality approaches 58.7%, mainly due to metastatic spread and sepsis [4,5]. The poor prognosis might be due to the difficulty of identifying tumorous lesions within an area of HS. Furthermore, sinus tracts are thought to facilitate local dissemination. This allows for the development of multifocal and extensive lesions hidden below the chronic scarring and nodules [5]. The management of cSCC in HS has not yet been standardized, but large surgical excision with at least 2 cm margins associated with radiotherapy appear to be the best therapeutic options [3,4]. TNF-α inhibitors, a therapeutic option that has been increasingly used for refractory cases of HS, have also been proposed as a contributory factor for

both development and aggressiveness of theses tumors [5]. The development of cSCC is one of the most dreaded complications of severe, long-standing HS [6]. Physicians should ensure a regular follow-up of longstanding HS lesions is crucial for timely diagnosis and appropriate surgical treatment [4].

5. Acknowledgment

None.

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